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Contents

1. [Editorial](#)
2. [Hair dye – an emerging suicidal agent our experience](#)
3. [Giant Cystic Hygroma colii in an adult a rare case report](#)
4. [Dual ectopic thyroid a case report of a very rare disease](#)
5. [Hypoplasia of paranasal sinuses case series with review of literature](#)
6. [Elongated styloid process a case report](#)
7. [Mucormycosis a case series](#)
8. [Letters to editor](#)



Otolaryngology online journal

Editorial

ARE YOU GEARING UP FOR THE Oto-rhino-laryngological REVOLUTION?

Down the memory lane, if you look back, the ENT doctor used to carry ear syringe, torch or head mirror, wax hook and the tonsillectomy set, that is what they were famous in those days.

Later, when compared to their counter-part (General Surgeons) ENT surgeons were having (in those days) sophisticated gadgets like auriscope, audiometer and operating microscope.

Recently, we hardly see any Otorhinolaryngologist without Endoscopes of one sort or other and the micro-debrider. We do have most of the recently developed gadgets put into use for various applications in our field, like Laser, Co ablator, Harmonic scalpel etc. Thanks to these improvements which made invisible areas from blind surgical manipulation to precision and targeted operative treatment.

We have widened the scope of ENT surgery into Head and Neck Surgery as well as Skull Base surgery, of course as a team work along with Neurosurgeons, making inaccessible areas to be managed with surgery.

Future is having much more in store. Medicine in general, for that matter Otorhinolaryngology field in particular is fast growing in the adaptations of newer sophistications and innovations.

To begin with, let me put forward that nanotechnology is going to rule the future world and this application is going to have tremendous influence in the life style of mankind. This nanorevolution is going to improve all the fields, which will be reflected favorably as well as adversely. Hence, let me caution that all Oto-rhino-laryngologists should keep updated so that they should not become “nano- illiterates”. There will be many regulations to monitor the usage of nanotechnology.

To expect a few advances like the drug delivery system, genetically favorable drugs, drugs to

modify genetic sequence for medical usage, remote controlled monitoring and management of illness, microbots for remote controlled micro surgery, miniature devices, miniature surveillance systems as well as alternate energy source and economical usage of power for effective and efficient control of wastages are going to be much more useful for effective management of illness. Further they are going to reduce co morbidities and the therapy as such is going to be more acceptable and pleasant.

Miniature cochlear implant, weight less or weight reduced BAHA, miniature stapler and micro glues on the one side and tissue banks tissue culture centers as well as more acceptable implant materials and biologically similar organ mimics are likely on the pipeline. Gene therapy and stem cell manipulations are useful for deafness and other genetic disorders.

Lastly, but not the least, let me say that Surgery comes to the rescue when there is no answer with medicine. Sooner we are going to observe the answers effectively. More and more conditions are likely to be treated with medicine. Surgical techniques will be modernized to suit the fast moving modern world, reducing convalescence period for early ambulance and rejoin the routine.

Dr. U. Venkatesan.



HAIR DYE- AN EMERGING SUICIDAL AGENT: OUR EXPERIENCE

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Abstract:

Hair dye poisoning has been emerging as one of the important causes of intentional deliberate self harm. To study the epidemiology of patients who consumed Super Vasmol (a commonly used commercial hair dye), profile of patients referred to the ENT department, to find the effectiveness of supportive therapy and to compare between the tracheostomy and the non tracheostomy group, a retrospective study was conducted in patients who consumed Super Vasmol admitted over a year (April 2009 – March 2010) in a teaching hospital in South India. Out of the 108 patients 38 (35.2%) were males and 70 (64.8%) were females. 42 (38.9%) were in 21-25 age group. Patients who developed cervico-facial oedema, throat pain, burning sensation in the throat, change of voice, no voice and dyspnoea with stridor were referred to the ENT Department. 74 (68.5%) patients needed an ENT opinion. Cervico-facial oedema was the main clinical manifestation in 74 patients. 33 of them underwent tracheostomy. The mortality in 108 patients was 22.2%

and after tracheostomy 21.2%. Purpose of the study is to create public awareness about the lethal effects of the hair dye, because it is a major health problem and to educate the public and the medical professional about the need for aggressive and early treatment.

Keywords:

Super Vasmol, cervico-facial oedema, Dyspnoea, Stridor, Tracheostomy, Paraphenylene diamine.

Introduction:

Suicide was responsible for about 6,00,000 deaths in the 1990's [1]. It is ranked as the third leading cause of death in the age group 15-44 years. Suicide rates have increased by 60% in the past 50 years [2]. Hair dye and its constituents paraphenylene diamine have been reported as an accidental and intentional cause of poisoning from developing countries in Africa [3,4,5,6] and Asia [7,8,9,10,11]. Case series have been reported from Khartoum [12,13,14], Casablanca [15] and Morocco [16]. Numerous papers have been published from India [8,9,10,11,17,18,19,20,21]. In India it is reported from Andhra Pradesh [9,10] and from Tamil Nadu (Vellore) [8,21]. It is a cheap, freely available emulsion based hair dye used for deliberate self harm in India [22]. Super Vasmol poisoning is being reported from developing countries probably due to its easy availability and its low cost [23].

It contains potential toxins including paraphenylene diamine, resorcinol, sodium ethylene diamine tetra acetic acid (EDTA), liquid paraffin, propylene glycol cetostearyl alcohol, sodium lauryl sulphate, herbal extracts, preservatives and perfumes [21,24]. The characteristic triad of features encountered are early angioneurotic edema with stridor, rhabdomyolysis with chocolate colored urine and acute renal failure [2]. Whenever this combination occurs, hair dye poisoning is a strong suspect. The combined effects of the individual toxicants results in significant morbidity and mortality [21].

Methods and materials:

All patients who consumed super Vasmol admitted in our hospital from April 2009 to March 2010 were included in our study. The total number of patients were 111. Two patients more than 55 years of age and one patient who arrived late (>48 hrs) to the hospital were excluded from the study. Hence, 108 patients were included in our study. The data were collected in relation to age, sex, amount of poisoning, time to reach the hospital, time of development of cervico-facial edema and ENT manifestations. All the patients were treated symptomatically with antihistamines, steroids, antibiotics, adrenaline nebulisation, nasal oxygen and forced alkaline diuresis. Patients who developed cervico-facial oedema, throat pain, burning sensation in the throat, change of voice and no voice were referred for an ENT opinion. Tracheostomy was done for severe tongue oedema, sublingual oedema, submandibular oedema with stridor. We have compared the profiles between alive and dead patients after the tracheostomy (Group A), alive and dead patients who have not undergone tracheostomy (Group B), and those who have and have not undergone tracheostomy (Group C). Analysis was done with statistical package for social sciences (SPSS v.15). Chi square test was used to evaluate the level of significance and P value < 0.05 was considered as significant.

Results:

The total number of patients included in our study was 108. The age group ranged from 13-45 with a mean age of 24.7 (SD- standard deviation 6.51). Among them 38 were males (35.2%) and 70 were (64.8%) females, with a male female ratio of 1:1.84 (Table 1). The female patients were predominantly working in small-scale industries in nearby places (45), some were house wives (40) and some were studying in schools (23). The age group ranged from 10 -55 years with more patients in the 21-25 years group (Table 2).

85% of patients told that they have chosen Super Vasmol as a poison because it is easily available, applicable and cheaper than pesticides. 90% of the patients had ingested the dye as an act of deliberate self harm. 10% of them had ingested to threaten their relatives without knowing the seriousness of the poison. 62% of the patients told that they got the information of Super Vasmol poisoning from the neighbourhood and 38% told that they got their information from the media.

The mean amount of ingestion was 75- 100ml. There is no apparent dose-toxicity relationship. The mean time of arrival to the hospital was ranging from 1hr and 30 min to 10 hrs with mean of 4 hrs 63 min (SD 1.73). The time of development of cervico-facial oedema ranges from 1hour and 30 minutes to 9 hrs with the mean time of 4 hrs and 29 min (SD 1.75).

Among the ENT manifestations there is a high incidence of development of cervico facial oedema (74 patients) followed by throat pain (48), stridor (33), change of voice (31) and no voice (12) in various combinations (Table 3). Of all these, the earliest sign is development of submandibular oedema. 33 of them developed dyspnoea with stridor and

needed emergency tracheostomy, because they did not respond to antihistamines, steroids and adrenalin nebulisation. Among the patients who underwent tracheostomy 6 (18.2%) were males and 27 (81.8%) were females. Among the patients who underwent tracheostomy 5 (4.6%) of them had complications. Three of them had tracheo-esophageal fistula and two had wound infection around the tracheostome and all of them were treated. But none of them died. 7 patients out of the 33 died due to other complications of the dye and not due to tracheostomy. Among the patients who died after tracheostomy 1 patient (14.3%) was male and 6 (85.7%) were females. Out of 108 the patients 24 (22.2%) patients died. 33 (30.5%) patients needed tracheostomy.

Table 1 Showing sex distribution (n=108)

| Sex | Total number of patients |
|--------|--------------------------|
| Male | 38(35.2%) |
| Female | 70(64.8%) |

Statistical comparison of age of patients who were alive (23.5) and died (30.4) after tracheostomy (Group A) showed a P value of 0.005, which is statistically significant (Table 4). The time of development of cervico-facial edema between alive (5hrs and 8min) and dead (2hrs and 58min) shows statistical significance (P value 0.001). There is no statistical significance between these groups in time to reach the hospital and the amount of poisoning. During the analysis of the non tracheostomy group (Group B) statistical significance is noted in age of the patients who were alive (26.03) and dead (19.8) with a P value of 0.001. There is no statistical significance in the profile of patients who have and have not undergone tracheostomy (Group C). The proportion of mortality in 108 patients was 22.2%, after tracheostomy 21.2% and in the non tracheostomy patients 22.7% (Table5).

Table 2 :Showing age distribution (n=108)

| Age group | Total number of patients |
|-------------|--------------------------|
| 10-15 Years | 5(4.6%) |
| 16-20 Years | 22(20.4%) |
| 21-25 Years | 42(38.9%) |
| 26-30 Years | 16(14.8%) |
| 31-35 Years | 15(13.9%) |
| 36-40 Years | 5(4.6%) |
| 41-55 Years | 3(2.8%) |

Table 3 Showing ENT manifestations

| Symptoms | Number of patients |
|----------------------|--------------------|
| Cervicofacial oedema | 74(68.5%) |
| Throat pain | 48(44.4%) |
| Stridor | 33(30.5%) |
| Change of voice | 31(28.7%) |
| No voice | 12(11.1%) |

Table 4 Showing comparative trends between A,B,C groups

| | A | | | B | | | C | | |
|--|------|-------|---------|-------|-------|---------|-------|-------|---------|
| | A1 | A2 | P Value | B1 | B2 | P Value | C1 | C2 | P Value |
| Number of patients | 26 | 7 | - | 58 | 17 | - | 33 | 75 | - |
| Age (Years) | 23.5 | 30.6 | 0.005* | 26.03 | 19.88 | 0.001* | 25 | 24.6 | 0.795 |
| Time to reach Hospital (hours) | 5.27 | 5.1 | 0.735 | 4.37 | 5.16 | 0.999 | 5.21 | 4.55 | 0.469 |
| Time of development of symptoms(hours) | 5.05 | 2.58 | 0.001* | 4.29 | 4.45 | 0.779 | 4.21 | 4.34 | 0.734 |
| Amount of poisoning (ml) | 92.5 | 119.2 | 0.172 | 98.17 | 98.18 | 0.843 | 98.18 | 98.18 | 0.999 |

* P value < 0.05 statistically significant

A group : Alive (A1) and dead (A2) patients after tracheostomy

B Group: Alive (B1) and dead (B2) patients who have not undergone tracheostomy

C Group : Patient who have undergone tracheostomy (C1) and have not undergone tracheostomy(C2)

Table: 5 Showing proportion of mortality

| | Total no of patients | No of deaths | Percentage |
|------------------|----------------------|--------------|------------|
| Tracheostomy | 33 | 7 | 22.70% |
| Non-tracheostomy | 75 | 17 | 21.20% |
| Total | 108 | 24 | 22.20% |

Discussion:

The first artificial dye was synthesised in the laboratory in 1856. Permanent hair colorants have been in commercial use for over 100 years [25]. Paraphenylenediamine is a coal tar (Paranitroaniline) derivative which, on oxidation produces Bondrowski's base, which is allergenic, mutagenic and highly toxic [2] and is used to shorten the duration of applications as well as intensify the color of henna (*Lawsonia Alba*) which is traditionally used to color the palms and hair [5]. Nott first documented paraphenylenediamine poisoning in a owner of a hair salon in 1924 [4,26].

Paraphenylenediamine causes severe angioneurotic edema, muscular edema, shock, rhabdomyolysis and intravascular haemolysis with haemoglobinuria (chocolate colored urine) culminating in acute renal failure [2,11], oliguria, acute tubular [12], focal glomerular sclerosis [5] cardio toxicity resulting in myocarditis [2] and fatal arrhythmias [5] causing sudden death. It causes rhabdomyolysis by promoting leakage of calcium ions from

The smooth endoplasmic reticulum which results in prolonged muscle contraction and irreversible change in muscle structure [22]. Other features include anemia, leukocytosis, liver necrosis [27], metabolic acidosis and hyperkalemia [9]. Cetostearyl alcohol, the combination of aliphatic alcohol and esters acts as a non-ionic surfactant and can produce allergic and urticarial reactions and sodium lauryl sulphate causes irritation to the skin [24]. Resorcinol a phenolic derivative, corrosive chemical used in hair dye, tanning and photography is neurotoxic, causing seizure, lethargy, coma and death [21] and is also nephrotoxic [2]. It can also cause nausea, dyspnoea, hypotension, diaphoresis, salivation, meth haemoglobinemia, bronchospasm and in severe cases pulmonary edema [9]. EDTA produces headache, vomiting, gastritis [24]. Propylene glycol is a viscous, colorless solvent is a potent nephrotoxic. It causes hyperosmolarity, raised anion gap, metabolic acidosis, Central nervous system depression, arrhythmias and renal dysfunction [2,29] and acute haemolysis [24]. Reddy I S et al reported nephrogenic systemic fibrosis causing bilateral symmetrical thickening and induration of skin over the extremities, verrucous papules and plaques following hair dye ingestion induced acute renal failure [30].

Hair dye poisoning has been reported in 4 female patients from Casablanca [15]. Yet another two papers show this poisoning in young girls [9,24]. A study by Anugrah Chrispal et al shows (11 out of 13) female predominance [21]. In

our study females contribute to 64.8% (70 out of 108) with the male female ratio of 1:1.84. The female patients were predominantly working in small-scale industries in nearby places (45), some were house wives (40) and some were studying in schools (23).

A number of children have been intoxicated in Sudan [14]. 6 year old child with paraphenyline diamine intoxication has been reported from Israel [31]. Bourquia et al has reported 4 patients in the age group between 18- 35 [15]. Another study shows the mean age as 27.75 [21]. A study of 374 patients of paraphenylene diamine poisoning from Morocco showed 54% were in 15-24 yrs and 11.5% were children [4]. In our study the age group ranged from 13-45, with the mean age as 24.75. The maximum number of patients (42) were in the 21-25 age group followed by 16-20 age group (22). Love affairs, dowry, divorce, illegitimate pregnancy, extramarital affairs and family conflicts play a critical role in this age group.

The mean amount of poisoning in Bhargava et al study was 300ml [8], in Anugrah Chrispal et al study was 150 ml [21], in Verma et al study was 100 ml [9] and in our study it is 75-100 ml. There is no apparent dose toxicity relationship noted.

The mean time to reach the hospital was 32 hrs 7 min in a study [21] 8 hrs in a case report by Dr. B.K. Barik et al [24] and 1 hr in Verma et al study [9]. In our study the mean time to reach the hospital was 5 hrs 3 min with a range from 1hr 30 min to 10 hrs. Time delay was due to the relatives not knowing immediately that the patient had consumed the poison or the distance from the hospital or transportation problem. The first clinical manifestation (usually cervicofacial edema) following ingestion of Super Vasmol occurs mostly within 6 hrs [5]. We have observed that the time to develop cervico-facial edema ranges from 1 hr 30 min to 9 hrs with a mean time 4hrs 29min (SD 1.67).

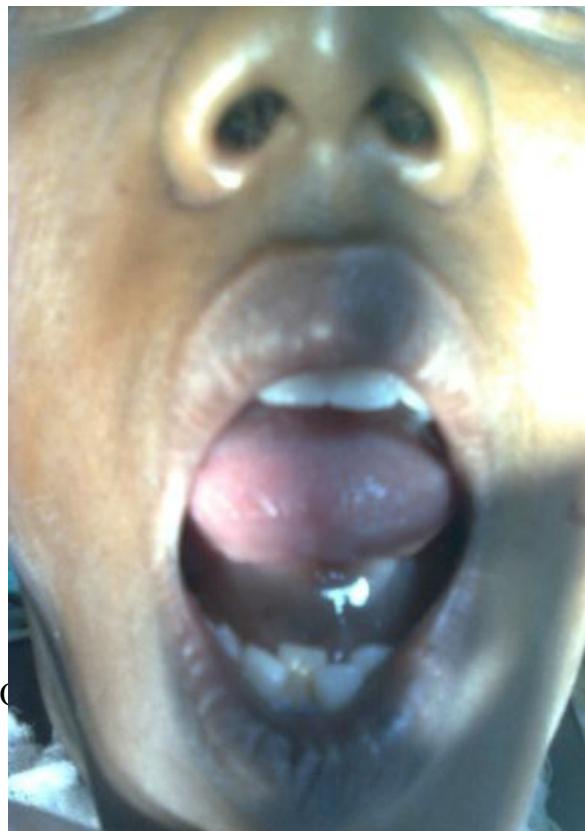
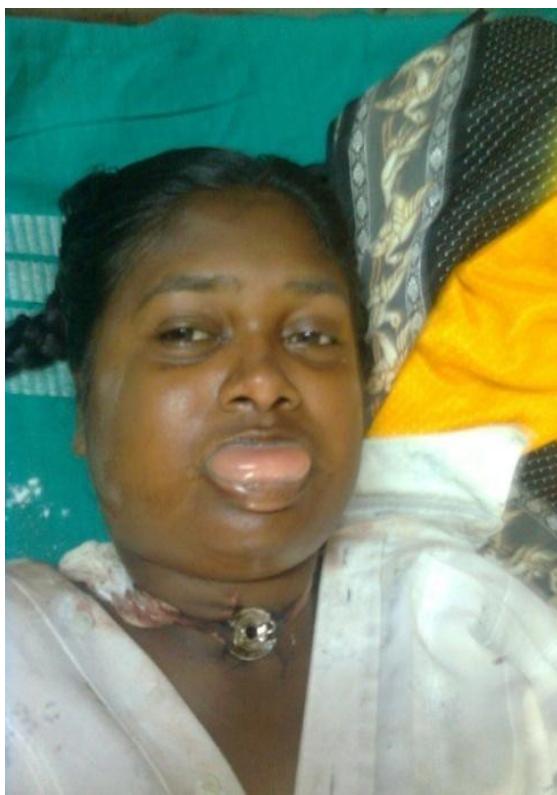
Apart from respiratory, renal, neurological, haematological [9] and cardiac manifestations[2,5] were also noted in Super Vasmol poisoning. Respiratory syndrome following Super Vasmol ingestion is represented by asphyxia and respiratory failure secondary to inflammatory oedema involving the cricopharynx and larynx [2]. Clinical manifestations resembling ludwigs angina was noted in one patient with sorethroat, cough, severe dyspnoea caused by edema of the tongue, pharynx and neck [31]. This compound can cause angioneurotic edema with respiratory distress [8, 14].

Swollen face with asphyxia was noted by Yagi et al [12] and Bourquia A et al [15]. Gross swelling of the neck and face below the chin and mandible on both sides was noted in Dr.B.K.Barik et al study [24]. Face and neck swelling with respiratory distress and convulsions was noted in a study by Verma et al [9]. Kallel et al conducted a study in 19 patients with paraphenylene diamine intoxication in Tunisia over 6 years and found that the predominant clinical features were cervico facial edema in 79% of the patients, upper airway tract edema in 68.4%, muscular edema in 26.3% and shock in 26.3% [6]. Suliman et al who studied 150 patients with paraphynele diamine poisoning found angioneurotic edema in 68% [6]. Anugrah Chrispal et al in their study with 13 patients with Super Vasmol poisoning found cervico-facial edema in 9 patients (69.2%), dyspnoea in 7 (53.8%) and inability to open the mouth in 6 (46.2%). [21]. In our study cervico- facial edema developed in 74 patients (68.5%), throat pain in 48 patients (44%), dyspnoea with stridor in 33 patients (30%), change of voice in 31 patients (28.7%) and aphonia (no voice) in 12 patients (11.1%). Whether cervico facial edema is a result of angioneurotic edema [5] or a result of direct injury and inflammation is unclear [21].

Hair dye poisoning is a medical emergency. Since there is no antidote, intensive supportive care is the cornerstone of management [21]. Treatment is mainly supportive with parenteral corticosteroids and antihistaminics have been tried. But controlled studies are needed [27].There are no therapeutic trials as to their benefit [21]. Calcium gluconate has been used to treat symptomatic hypocalcemia causing tetany and seizures [8]. Metabolic acidosis should be corrected [2]. Early intervention with half normal saline and sodium bicarbonate infusion have been shown to have beneficial results in rhabdomyolysis and helps in avoiding dialysis [32]. Dialysis per se is supportive as the toxin is not dialyzable [7,27].

Yagi et al found intensive medical treatment along with tracheostomy as life saving [12]. In Ram et al series 4 patients had asphyxia and cervico facial edema resulting in emergency tracheostomy [18] and in Suliman et al studies it is 15.8% [5]. Ventilatory support was needed for a patient [9]. Patients should be monitored for respiratory distress and endotracheal intubation has to be performed early if laryngeal edema develops [2]. We treated our patients with parenteral Hydrocortisone, Chlorpheneramine maleate,,Cefotaxime, Ranitidine, Adrenaline nebulisation, nasal oxygen and forced alkaline diuresis. Tracheosomy is done if the cervico-facial edema is severe and causes stridor and in those who developed respiratory distress. In our series 74 patients developed cervico-facial edema and 33 patients required emergency tracheostomy.

Morbidity and mortality rates are high once renal failure develops. Mortality rates vary between 0.03%- 60% [2]. High mortality with in 24 hrs was noted in Sir Hashim et al study 41% (13 out of 31) [14]. In Ram et al study it is 21% [18]. In another study by Mohamed Abdelraheem et al the mortality rate is 12-42% [33]. In our study, the propotion of mortality in 108 patients is 24 (22.2 %), in tracheosomty patients 7 (21.2%) and in non tracheostomy patients 17 (22.7%). There is no significant difference in mortality in these groups. When we consider the whole population the mortality of Super Vasmol is 1.38/ 100,000.



Pictures showing lingual and sublingual oedema

Conclusion:

There is a high incidence and rapid rise of Super Vasmol hair dye poisoning in this area. In our study age of the patient and the time to reach the hospital and time of development of cervico facial edema have an impact on mortality. Young adults (of the age group 21-25) are at the highest risk. The amount of poison ingested and tracheostomy do not have a significant impact on mortality. Many of them develop severe cervicofacial edema, dyspnoea with stridor within a few hours of consumption. So, reaching the hospital promptly and timely medical help has a major impact on mortality. Many patients also developed renal failure and cardiac arrhythmias. These alarming facts suggest the necessity to impose regulation on the free availability of Super Vasmol hair dye. Proper toxicity warning on the hair dye labels, counselling adolescent girls to withstand stress, public education regarding the potential toxicities of these compounds are imperative to reduce the mortality. It is also important for the medical fraternity to know about the manifestations, early detection and prompt treatment of the patient.

Key message:

Clinical outcomes rely on early recognition, prompt referral and aggressive treatment in collaboration with different specialities. Awareness programs about its toxicity should be implemented at different levels.

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Conflict of interest: None

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Otolaryngology online journal

GIANT CYSTIC HYGROMA COLLI IN AN ADULT- A RARE CASE REPORT

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INTRODUCTION

Cystic hygroma (lymphangioma) is often a benign congenital malformation of the lymphatic system that occurs as a result of sequestration or obstruction of lymphatic vessels. These lesions are usually discovered in infant or children younger than two years of age. Occurrence in adults is uncommon, and fewer than 100 cases of adult lymphangioma have been reported in the literature^(1,2). The objectives of this case report are to present the clinical history and surgical findings, review of literature and the unique problems encountered in the surgical management of this particular patient.

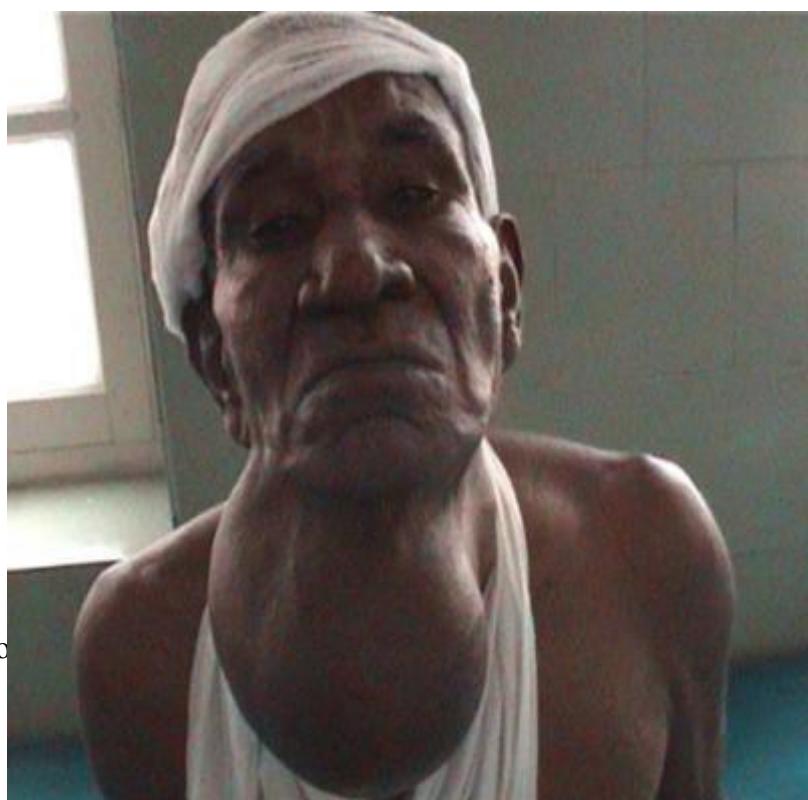
Case report

HISTORY:

A 75 yr old south Indian male, an agricultural labourer presented in our OPD with complaints of swelling in the neck of 2 years duration. It was slowly increasing in size and posed a restriction to neck movements. He had no complaints regarding his voice or deglutition..

He was particularly alarmed and concerned about the fact that following aspiration of the swelling a couple of weeks ago by a private practitioner the swelling had reappeared with increased vigour and attained its original size. He was not a diabetic and had no history of trauma or respiratory tract infection in the recent relevant past.

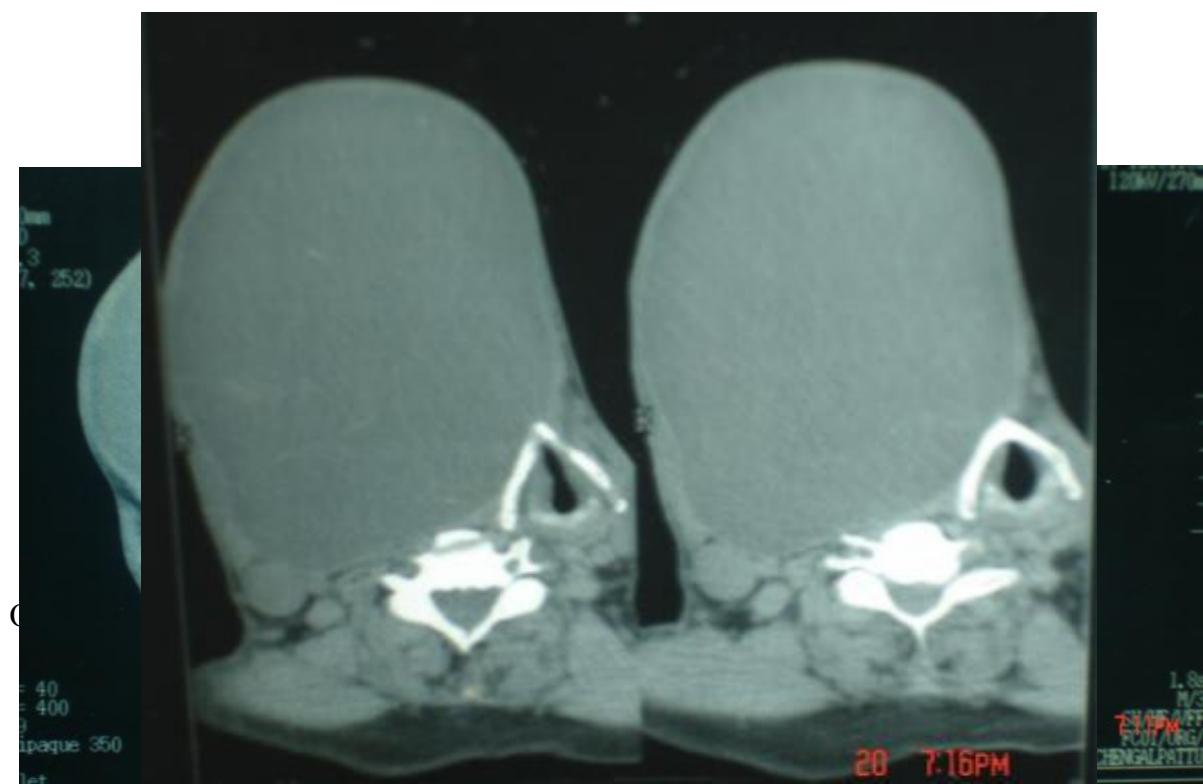
Physical examination revealed a diffuse, smooth, soft fluctuant non transilluminant swelling measuring 20 /18 cm which occupied the whole of the anterior neck pushing the right sternocleidomastoid muscle and carotid laterally. The swelling was predominantly present over the right side and encompassed the submandibular, carotid, muscular and posterior triangles on the right side. Indirect laryngeal examination revealed normal mobile vocal cords.



On the left side it did not cross lateral to the sternomastoid muscle .the larynx and trachea were not palpable in the midline and were pushed far to the left and were palpable along the lateral border of the neck on the left. The left carotid was palpable in normal position and was not displaced. A presumptive diagnosis of a cystic hygroma was made and investigations were carried out.

IMAGING:

The contrast enhanced CT of the neck revealed a non enhancing, homogeneous soft tissue mass in the neck extending from the submandibular region to the supraclavicular fossa. The airway was pushed to the left and there was no retrosternal extension of the lesion. There was normal enhancement of the great vessels and were normal except for the lateral displacement.



Aspiration revealed serosanguinous fluid with normal epithelial cells and a few inflammatory cells. No malignant cells were seen.

SURGICAL TECHNIQUE:

This case posed unique problems because of the deviated airway. Intubation was done blindly over a bougie with a cuffed size 8 endotracheal tube. A transverse skin crease incision was made 5 cms above the suprasternal notch. Subcutaneous tissue was dissected and the platysma incised. The anterior jugular veins were encountered and ligated on both sides. The strap muscles were found to be stretched and thinned out over the swelling. While opening the strap muscles the swelling was inadvertently punctured and about 2 litres of serosanguinous fluid was let out. The cyst wall found to be thickened and contained follicles on the inner secretory surface. The cyst wall was completely dissected in toto from the surrounding structures and was found to be slightly adherent to the thyroid and cricoid cartilage. However a plane of cleavage allowed complete dissection from the cartilage. The hypoglossal nerve was encountered on the right during dissection and was preserved. There was no injury to any of the neurovascular structures of the neck. The excessively stretched out skin was trimmed and the wound was closed in layers with a drain.



There was a serous collection in the drain which continued for three days and eventually stopped. Tight compression dressing was made to avoid recollection. The patient was discharged on the 10th day without any complications. It has been one year following surgery and patient has had no recurrent swelling.



HISTOPATHOLOGY:

Histopathologic diagnosis was cystic lymphangioma which was supported with a thick fibrous wall,

containing dilated blood vessels partly filled with erythrocytes and infiltrated with lymphocytes.

DISCUSSION:

Cystic hygroma or giant lymphangioma is a benign malformation of the lymphatic system. It is believed to arise from a congenital malformation of the lymphatic system in which a failure of communication between the lymphatic and venous pathways leads to lymph accumulation¹. Most cystic hygromas present in-utero or in infancy and therefore most of the literature on management considers paediatric cases. The effect of these lesions depends on their position and relationship to surrounding structures. Although the lesion can occur anywhere, the most common sites are in the posterior triangle of the neck (75%), axilla (20%), mediastinum (5%), groin, retroperitoneal space and pelvis². To date there have been fewer than 150 reports of adult cervicofacial cystic hygroma in the English language literature and the optimum management of these lesions is still a matter of debate.. Thirty-two patients with cervical lymphangioma were treated at the Mayo Clinic; this is the largest series of the literature³. However, rapid enlargement over a short period of time has frequently been reported³ and major structures such as the larynx, trachea, oesophagus, brachial plexus and great vessels have known to be compressed or incorporated within the lesion. .. Diagnosis in adults is considered to present a greater challenge than in children and initial misdiagnosis, frequently as branchial cleft cysts¹.

There are three histological subtype. Capillary lymphangioma (composed of small lymphatics), cavernous lymphangioma (composed of larger lymphatics), cystic lymphangioma (cystic hygroma-composed of large macroscopic lymphatic spaces with collagen and smooth muscle)¹. Cavernous lymphangioma is the most common subtype. But cystic lymphangioma occurs approximately 1 in 12000 births and 95% occurring by the second year of life¹.

Cystic hygroma could be classified into septated (multiloculated) or non-septated single cavity (non-loculated). Presentation in adulthood is rare and the cause is uncertain, although trauma and upper respiratory tract infection have both been suggested as possible triggers for onset^[5,6]. In this case there was no identifiable cause and onset was sudden

Proposal for staging of lymphatic malformations of the head and neck (adapted from de Serres[8])

Stage Location of lesion

I Unilateral infrathyroid

II Unilateral suprathyroid

III Unilateral infrathyroid and suprathyroid

IV Bilateral suprathyroid

V Bilateral infrathyroid and suprathyroid

Our case belonged to stage v

Complete surgical excision has traditionally been considered the treatment of choice for cystic hygroma^[6]. However, several authors have suggested that sclerotherapy may be a more appropriate first-line therapy^[7]. Although sclerotherapy is now well established in the treatment of neonatal and

paediatric cystic hygromas, there have been relatively few cases reported of its use in adult patients. Some success has been reported in small numbers of adults with sclerotherapy agents such as OK-432[7]. Caution has been urged with the use of agents such as OK-432 which induce a local immune response that often results in a rapid temporary increase in the size of the cystic hygroma[7]. Depending on the anatomical relations of the tumour, such increases in size may be intolerable and it has been suggested that such therapy should only be administered in specialised facilities due to the risk of airway obstruction[9].

Smith et al.[7] compared results from their large-scale trial of OK-432 with pooled results from large surgical case series reported in the literature and reported greater success rates and lower occurrence of major complications with OK-432 sclerotherapy compared with surgical excision. However, their study focussed mainly on children and only one of the comparative surgical cases included adult patients. Several authors have expressed the opinion that surgical excision of cystic hygroma is an easier procedure in adult patients, because these lesions are better circumscribed, and as such the success rate is greater[2,5].

In this case, it was thought that the ideal treatment would be complete surgical excision as a multiloculated cystic hygroma may not respond to sclerotherapy. Success of surgery has been found to correlate with histology, encapsulation, complete excision, anatomical location and stage of the lesion. Imaging appeared to show smooth margins, indicating a lack of infiltration, which is a good prognostic feature, facilitating complete removal and low recurrence. However, this was an extensive stage V lesion with close relations to major structures and therefore a difficult procedure was anticipated. It proved to be impossible to remove the cystic hygroma completely without rupture, a recognised problem as these tumours usually have a fragile thin wall. Intra-operative rupture of the lesion complicates complete removal as it obscures the limits of the structure[1]. However, Riechelmann reported very low levels of recurrence (1/9 patients) following subtotal excision when small plaques of tumour wall were known to be left in situ. To the best of our knowledge we were able to remove the cystic hygroma completely. Complications of surgery should be discussed with the patient before consent is obtained for surgery, including

-Difficult intubation

_ Scar: there may be a long scar depending on the size of the cystic mass but this should usually improve over time.

_ Injury to important structures and nerves: all blood vessels and nerves located between the mandible and the sternomastoid muscle are vulnerable to injury, specifically the mandibular branch of the facial nerve, the spinal branch of the accessory nerve and the greater auricular nerves and hypoglossal nerves.

_ Venous bleeding is a possibility but injury to the carotid sheath and its content and some external carotid branches should be rare.

_ Wound Infection.

_ Recurrence after surgery is a possibility.

CONCLUSION:

_ Cystic hygroma are congenital neck masses usually presenting at birth but may present at any age
_ Cystic hygroma is a rare presentation in adults

_ Malignancy should be excluded in all adult patients presenting with a cystic neck swelling
Adult-onset cystic hygroma

Differential diagnoses for a congenital neck mass in adults

- Branchial cleft cyst (lateral to midline)
- Dermoid cyst (midline)

- Thyroglossal cyst (midline)
 - Haemangioma
 - Thymic cyst (midline/lateral)
 - Neck malignancy (midline/lateral)
- Investigations should include:
- Ultrasound scan
 - Fine-needle aspiration (important to exclude malignancy)
 - Computed tomography or Magnetic resonance imaging
 - _ Final diagnosis depends on tissue analysis

Treatment options in adults include:

- Surgery
- Sclerotherapy
- _ In multiloculated cystic hygromas, surgery may be the preferred option but it is important that surgical complications are kept to a minimum.

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Dual ectopic thyroid – A case report of a very rare disease

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Introduction:

Ectopic thyroid is a developmental defect of thyroid gland that leads to presence of thyroid tissue at sites other than its normal pretracheal location. It is frequently found along the course of the thyroglossal duct but can also be found at remote distant sites. Lingual thyroid is the most common presentation of ectopic thyroid . It is very rare to have two ectopic foci of thyroid tissue simultaneously and only 19 cases of dual ectopia have been reported in world literature. In 70% of cases of ectopic thyroid the normal thyroid gland is absent. We report a case of 12 year old girl presented with dual ectopic thyroid.

Case report:

A twelve year old girl presented with history of a swelling in front of neck in the subhyoid area first noticed about two months back. The swelling gradually increased in size. There was no history of pain, or any pressure symptoms like dyspnoea dysphagia or dysphonias. Birth history ,developmental milestones were normal and menarche was not attained. A detailed general and systemic examination did not reveal any abnormality. Local examination revealed a 2.5 * 2 cm firm swelling in the subhyoid area and 2 * 2 cm midline swelling in the posterior part of tongue. Thyroid gland was not palpable at its normal site. Thyroid function test suggested primary hypothyroidism with elevated TSH , low T3 & T4 . Fine needle aspiration revealed colloid goitre. Ultrasonogram and CECT of the neck revealed absence of normally located thyroid and presence of dual ectopic thyroid – lingual and subhyoid.

Discussion:

Ectopic thyroid was first described by Hickman in 1869 in a newborn who was suffocated 16 hours after birth because of a lingual thyroid causing upper airway obstruction. Lingual thyroid is the most common ectopic thyroid accounting for 90% of all cases with a prevalence between 1: 100000 and 1:300000 and a clinical incidence between 1:4000 to 1:10000. Other sites of ectopic thyroid are suprathyroid and infrathyroid, lateral aberrant thyroid,

substernal goiters, struma ovary and struma cordis. Ectopic thyroid has also been found in larynx, trachea, oesophagus, pericardium, diaphragm and branchial cysts. Rare cases of ectopic thyroid are described in parathyroid, cervical lymph nodes, submandibular gland, duodenal mesentery, adrenals and carotid bifurcation. Ectopic thyroid occurs more commonly in females and are usually seen during adolescence and pregnancy when the demand for thyroid hormone increases. Up to 70% of patients with lingual thyroid had hypothyroidism and 10% suffer from cretinism. Rest are euthyroid. However hyperthyroidism has been rarely described. Our patient presented during adolescence with hypothyroidism. The thyroid gland is not found in its usual location in 70% of patients with ectopic thyroid. Ectopic thyroid is rare and is often mistaken for thyroglossal duct cyst.

Let's go through the development of thyroid briefly; In the third to fourth weeks of embryonic life, the thyroid gland appears as a midline diverticulum in the pharyngeal wall between the first and second branchial arches. The anterior two thirds of the tongue develops from the tuberculum impar while the posterior one third develops from the hypobranchial eminence and the junction forms the future foramen caecum from which the thyroid gland develops. The thyroid diverticulum becomes bilobed and descends in the neck and fuses with the 2 lateral diverticulae that are derived from the fourth pharyngeal pouch, which contributes to the parafollicular cells. As the developing thyroid advances caudally, it remains attached to pharyngeal wall by the thyroglossal duct, which usually obliterates on the sixth - to eighth weeks of life. The thyroglossal duct descends inferior to the foregut, passing in front of the hyoid bone and larynx and finally localizes in the lower neck anterior to the thyroid cartilage and first few tracheal rings. Ectopic thyroid occurs when there is an arrest or irregularity in such descent, the location of which decides the type of ectopia namely lingual (at the base of the tongue), sublingual (below the tongue), prelaryngeal (subhyoid) or substernal (mediastinal).

Presence of two ectopic foci of thyroid tissue simultaneously is rare and very few such cases of dual thyroid ectopia have been reported in world literature. In an extensive review of literature Sood et al found that the mean age of these patients was 15 years, more common in females with a F:M ratio of 1.25:1. The symptoms varied from asymptomatic to anterior neck swelling with or without altered thyroid status. In almost all of these patients one site of ectopy was at lingual or sublingual region. The second ectopic focus was at subhyoid or suprathyroid level in most cases.

Ectopic thyroid may be asymptomatic or produce symptoms due to its location. Lingual thyroid can cause foreign body sensation in tongue and dysphagia. Large blood vessels present on the surface of lingual thyroid predisposes it for ulceration. All diseases capable of affecting the normal thyroid can affect the ectopic thyroid like adenoma, hyperplasia, inflammation and rarely malignancy. The rate of malignant transformation in ectopic thyroid is no greater than in normally placed thyroid.

A neck ultrasound examination, neck CT scan are valuable modalities for forming a differential diagnosis. US examination plays a role in differentiating cystic and solid masses. On CT scan, thyroid tissue has a characteristically high enhancement due to its great vascularity. FNAC may exclude malignancy or other pathologic changes in heterogeneous tissue. Radionucleotide thyroid

scan can detect all the ectopic foci of thyroid in the body.

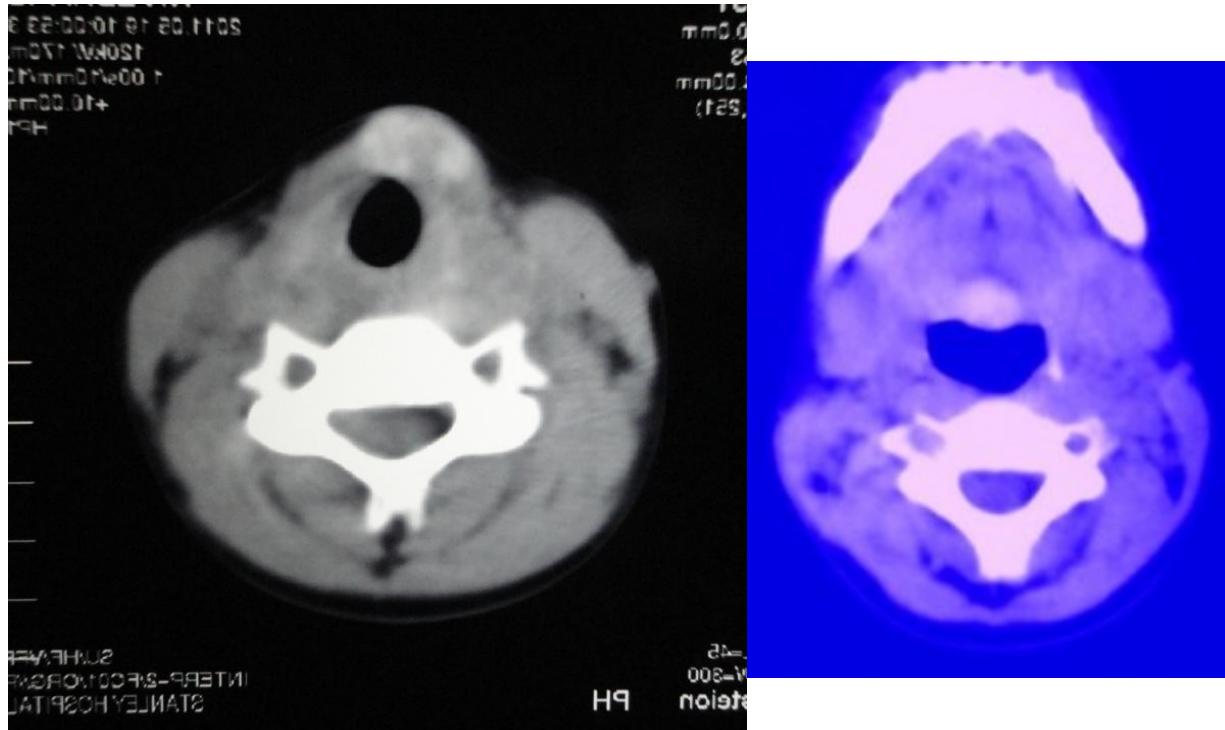
Asymptomatic and euthyroid patients do not require any treatment but they should be followed up and looked for any complications. Patients with raised TSH with swelling should be put on replacement therapy with thyroid hormone which can produce a slow reduction of the mass. When medical treatment fails or there are obstructive symptoms or haemorrhage or suspicion of malignancy then surgery should be considered. However, 70-90% of patients have no functioning thyroid tissue besides the ectopic thyroid, so extreme caution must be exercised; removal of the only functioning thyroid tissue will result in permanent hypothyroidism.

Our patient was put on 0.1 mg of levothyroxine once daily and on regular follow up for last 2 months. She attained menarche after 1 month of the thyroxine supplementation therapy.

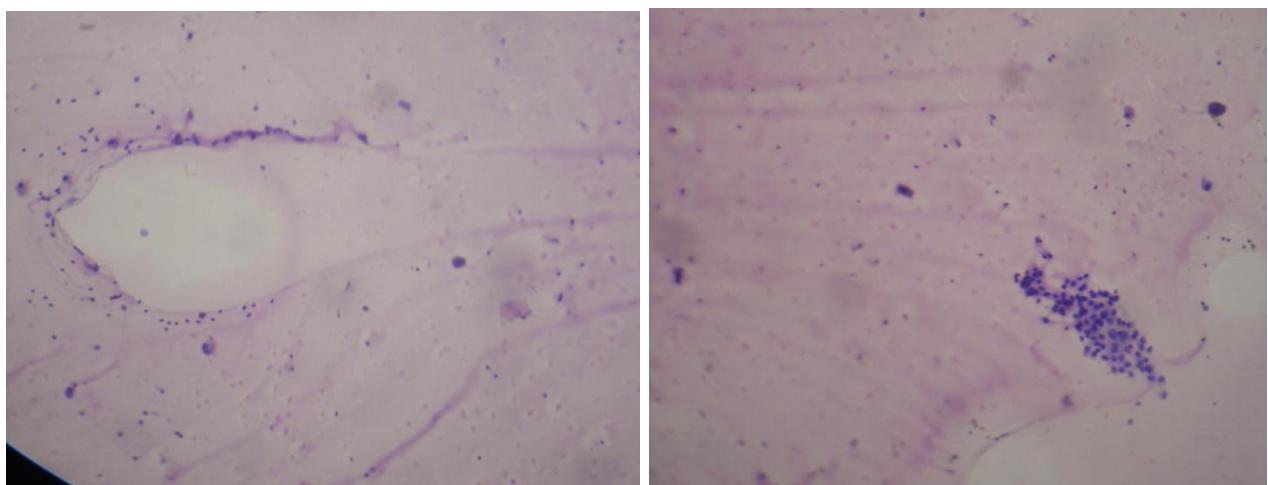
Conclusion:

Ectopic thyroid should be considered as a differential diagnosis in a case of midline neck swelling since it is often misdiagnosed as thyroglossal cyst. The possibility of another ectopic focus should be kept in mind when an ectopic thyroid is encountered. Unnecessary surgery must be avoided in case of asymptomatic ectopic thyroid as it may be the only functioning thyroid in the body in most of the cases.





CT scan showing subhyoid thyroid mass and lingual thyroid



FNAC showing colloid elements

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Bipul Kumar Choudhury, Uma Kaimal Saikia, Dipti Sarma, Mihir saikia



HYPOPLASIA OF ALL PARANASAL SINUSES A SERIES CASE REPORT AND LITERATURE REVIEW

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Abstract:

Hypoplasia of maxillary sinus is a rather rare condition. Review of literature reveal that so far only 6 cases have been reported. Hypoplasia of frontal and sphenoid sinuses has been documented rather frequently. In this article the authors report two rare cases of hypoplasia involving all paranasal sinuses which has not been reported so far in literature. This is actually an incidental finding when routine CT scan of sinuses was performed. This patient was clinically asymptomatic. Awareness of this condition is important because of implications involved in performing FESS surgery in these patients. Routine x-rays will lead to erroneous diagnosis of sinus infection because of the opacity seen in the poorly developed sinus area.

Introduction:

Hypoplasia of paranasal sinuses is rather rare. It can lead to problems in diagnosis, as they are commonly misdiagnosed as infections / neoplasm involving sinuses. Surgical attempts in these patients will be rather difficult and fraught with danger. Commonly reported hypoplasia involves maxillary sinuses. Incidence of maxillary sinus hypoplasia ranges between (1.5 – 10%)

Majority of these patients reported were asymptomatic and hypoplasia involving maxillary sinuses were identified only on routine radiology. Another study (2002) reports that only about 7 cases of true maxillary sinus hypoplasia have been reported.

Literature search did not revealed any report of Hypoplasia of all paranasal sinuses. These cases are being presented for their rarity.

Case Report I:

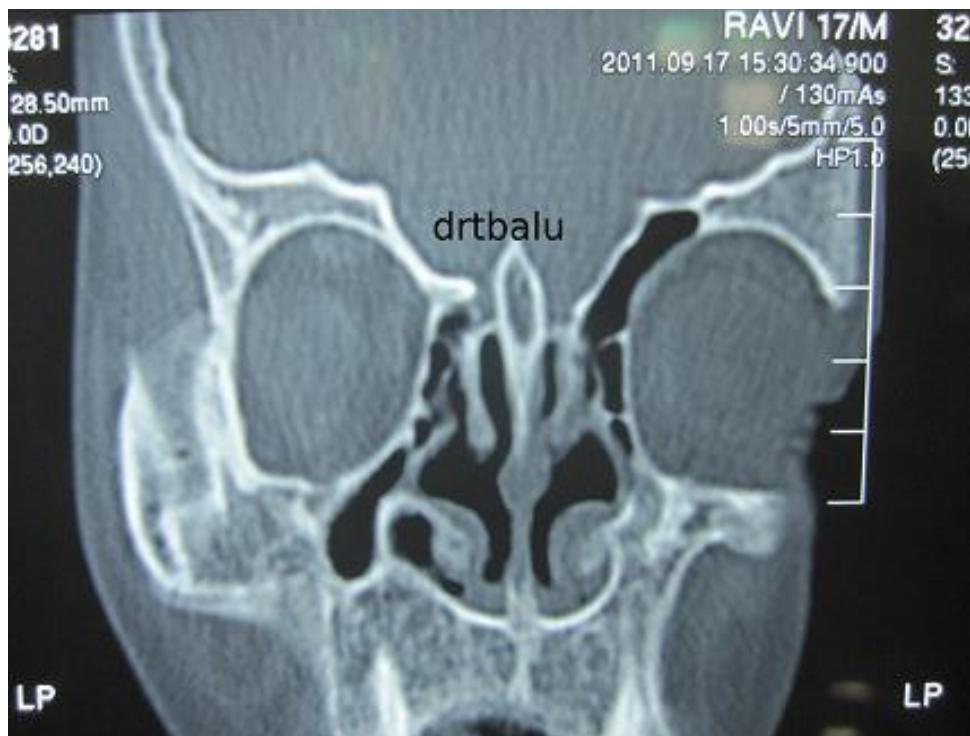
Female patient aged 30 reported to OPD for treatment of otitis media. She gave no history of head ache, discharge from nasal cavities. Clinical examination: Revealed a dry central perforation in her left ear. Clinical examination of the patient did not throw up any new findings. CT scan of nose and paranasal sinuses was taken to rule out focal sepsis in nose and paranasal sinus areas. This patient was actually asymptomatic.



Coronal CT anterior cut showing hypoplasia of frontal sinus



Coronal CT anterior cut showing hypoplastic frontal sinus



Coronal CT nose and sinuses showing hypoplastic maxillary and frontal sinus

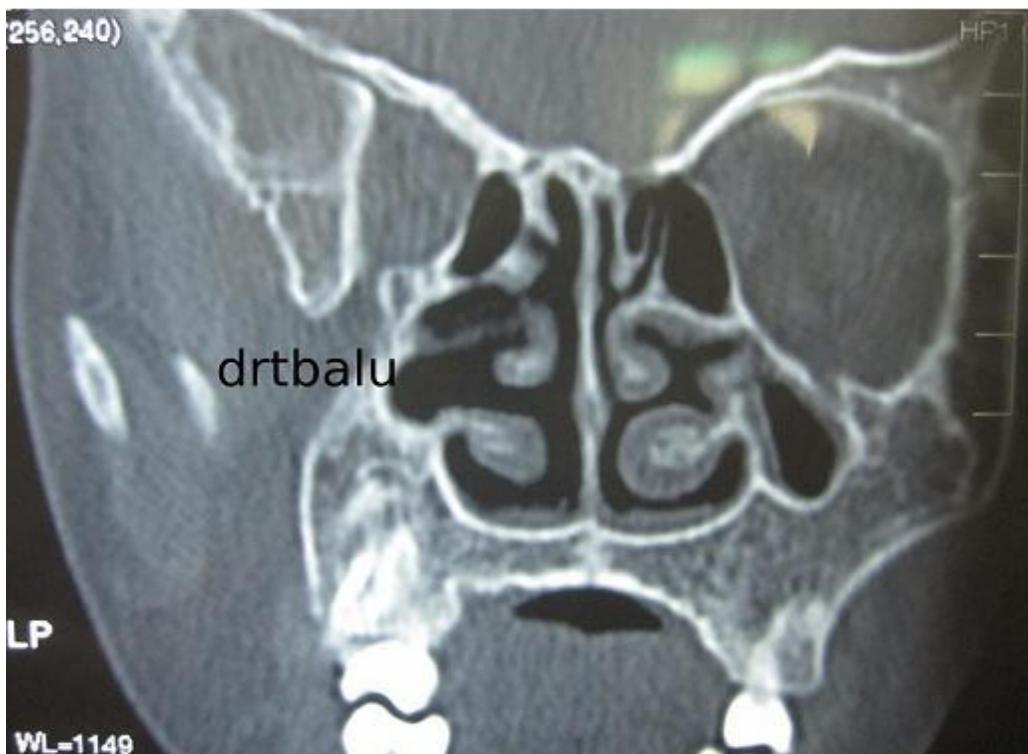
Case Report II:

30 years old male patient came to the hospital with H/O:

Watery rhinorrhea – 10 days Sneezing – 5 days

Anterior rhinoscopic examination revealed watery discharge from both nasal cavities. Right inferior turbinate showed evidence of mucosal oedema.

CT scan of nose and paranasal sinuses was performed as the patient insisted it.



Coronal CT nose and sinuses showing hypoplastic maxillary sinuses

Coronal CT scan performed in the patient revealed hypoplasia of both maxillary sinuses. Patient of course responded to conventional medical management.

Discussion:

An internet search revealed case reports involving only frontal, sphenoid and maxillary sinuses. They are all reported as individual isolated findings. Analysis of published literature revealed that hypoplasia is common in:

1. Frontal sinus
2. Sphenoid sinus
3. Rare in maxillary sinus

Maxillary sinus hypoplasia is rather very rare. Only 6 cases have been reported.

Reasons for maxillary sinus hypoplasia:

Hall's Hypothesis:

Hall proposed that intrauterine developmental anomalies to be the cause for hypoplastic maxillary sinus.

Wasson's Hypothesis: According to Wasson sinus infections during the first year of life could cause maxillary sinus hypoplasia⁴

Bolger's classifications of maxillary sinus hypoplasia:

Bolger was the first to associate hypoplasia / aplasia of uncinate process with hypoplasia of maxillary sinus. He considered ethmoid and maxillary sinuses to be intimately related embryologically. He suggested that developmental abnormalities involving uncinate process will lead to hypoplasia of maxillary sinus also. He classified hypoplasia of maxillary sinuses into three types.

Type I: Mild sinus hypoplasia. Normally developed uncinate process, well developed infundibulum with varying degrees of sinus mucosal thickening,

Type II: Significant maxillary sinus hypoplasia, hypoplastic / absent uncinate process, poorly defined or absent infundibulum, total opacification of affected sinus.

Type III: Profound hypoplasia of maxillary sinus, absent uncinate process.

Two case reports mentioned here belonged to Type II variety of Bolger.

Geraghty and Dolan 's diagnostic criteria for diagnosing hypoplasia of maxillary sinus:

1. Enlargement of vertical orbital dimension
2. Lateral position of infraorbital neurovascular canal
3. Elevated canine fossa
4. Enlargement of superior orbital fissure
5. Enlargement of pterygopalatine fissure

Bassiouny et al 's ,Classification of maxillary sinus hypoplasia:

They classified maxillary sinus hypoplasia into developmental and acquired categories.

Developmental categories:

1. Isolated hypoplasia due to developmental arrest due to infection / irradiation / injury
2. Developmental anomalies like facial dysostosis

Acquired categories:

1. Trauma with deformity due to fracture of facial skeleton / surgery
2. Inflammatory osteitis (Wegener's granuloma)
3. Hypoplasia due to Thalassaemia / Cretinism
4. Neoplastic osteitis

Conclusion:

Hypoplasia of maxillary sinus is very rare. It is asymptomatic and picked up as incidental finding in routine CT imaging. Hypoplasia involving more than one sinus is still rare. Awareness of this condition will help in avoiding complications liked missed ostia, breach of lamina papyracea during FESS surgery. It is always important to perform CT scan of paranasal sinuses both axial and coronal sections before performing FESS.

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ELONGATED STYLOID PROCESS-A CASE REPORT

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Abstract:

Elongated styloid process occur in 4 % of the general population, while only a small percentage of these patients are symptomatic. The normal adult styloid process length is considered to be between 20mm to 30mm. The longest recorded elongated styloid process so far that caused symptoms and hence underwent surgery was around 6.3cms though in an adult human dry skull upto 8cms have been recorded (2). We would like to present a case of an elongated styloid process measured 6.5cms which could be the longest recorded styloid process in world literature. A 45 years old female patient presented with persistent pain in the throat even after tonsillectomy done 4 years before. An elongated styloid process was diagnosed and was removed by transoral approach following which the patient was relieved from her symptoms.

Case Report:

A 45 years old female patient came with chief complaints of persistent pain in the throat that is in the left tonsillar fossa which gets aggravated on turning the head. She had undergone tonsillectomy 4 years before for recurrent throat pain. On clinical examination the elongated styloid process was palpated on the left tonsillar fossa .As the pain was persistent even after the surgery, radiological evaluations were made which revealed an elongated styloid process on the right side. Through an transoral approach the styloid process was removed which measured 6.5cms.This could be the longest recorded elongated styloid process recorded in the world literature in a live subject. Following surgery the patient was totally relieved of her symptoms.



Clinical photograph and CT scan of the patient

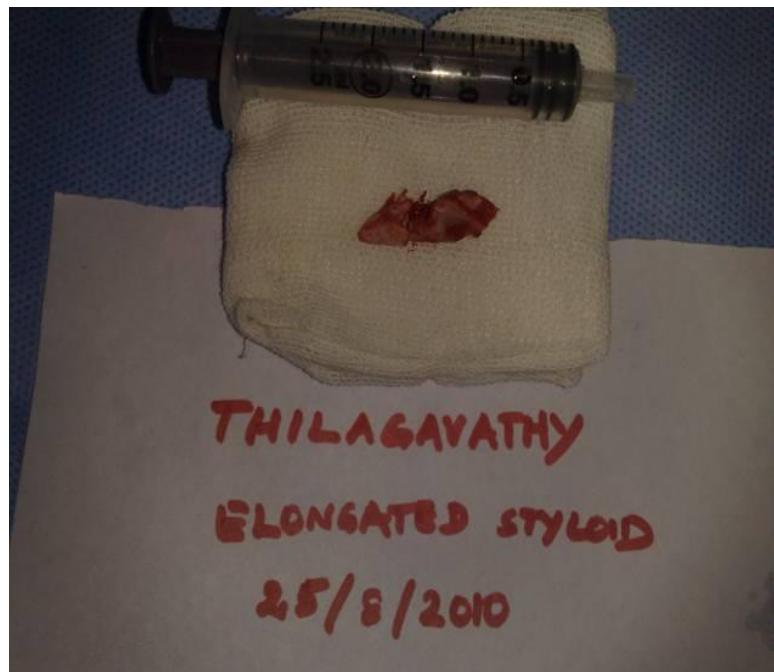


Figure showing specimen after removal

Discussion:

Styloid process of temporal bone is a slender projection attached to base of skull and the styloidhyoid apparatus extends from the tip of the styloid process, the stylohyoid ligament passes downwards and forwards to the lesser cornu of the hyoid bone. All these structures are derived from second bronchial arch cartilage⁽³⁾. Eagle defined the length of a normal styloid process at 2.5 to 3 cms. The normal length of styloid process varies greatly. An elongated styloid process occurs in 4% of general population which only small percentage (between 4 to 10.3%) of these patients are symptomatic. The styloid process and the styloidhyoid ligament have been linked to Eagle's syndrome which has a symptomatology characterized by the sensation of having a foreign body in the pharynx, causing difficult and painful swallowing and earache. It has been referred to as styloid syndrome, stylalgia, stylohyoid disorder, neuralgia of styloid process, cervicopharyngeal pain syndrome. Thot et al stressed that length in isolation is not a risk factor but that its combination with increased acuity in deviation from norm, both anteriorly and medially makes elongated styloid process the sole cause of Eagle syndrome⁽¹⁾. Diagnosis can be made by digital palpation of the styloid process in the tonsillar fossa⁽⁶⁾.

Diagnosis can be confirmed by imaging studies. Lateral view radiographs of the skull can be taken, but the disadvantage of this view is the overlapping between styloid processes of the both sides and with adjoining bone structures. An anteroposterior view radiograph should be taken to determine whether the styloid process is medially or laterally deviated. Orthopantomogram shows the entire length of the process distinctly and its deviation can also be made out clearly. CT scanning is extremely valuable tool. Treatment of symptomatic elongated styloid process includes both medical and surgical therapy⁽⁵⁾.

Medical management includes the following analgesics, anticonvulsants, antidepressant, local infiltration with steroids or long acting local anesthetic agents. Two surgical approaches to styloidectomy are the intraoral approach or (Transpharyngeal) and the extraoral approach⁽⁵⁾.

Main surgical complications associated with styloidectomy are deep space neck infection, injury to main neurovascular structures, haemorrhage, temporary alterations of speech and swallowing, injury of the facial nerve.

Conclusion:

Elongated styloid process is a diagnosis that should be considered in the evaluation of recurrent neck, throat or facial pain and dysphagia with or without radiation of pain to the ipsilateral ear⁽²⁾. Eagle's syndrome though the incidence is 4 to 7%, it is largely under diagnosed. A thorough clinical and radiological examination will reveal impending insult. Proper diagnosis can definitely be of immense help to rationalize the line of management and the ultimate clinical outcome⁽⁵⁾.

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Collar stud abscess an interesting case report

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Abstract – Cervical Lymphadenopathy with collar stud abscess of tuberculous etiology is uncommon nowadays. This case is being reported for clinical interest and for the purpose of documentation.

Introduction:

Tubercular Lymphadenopathy is a common extra pulmonary manifestation of tuberculosis. Collar stud abscess are rarely seen in OPD nowadays. Tuberculosis remains a problem throughout the world and is still a common cause of cervical lymphadenopathy.

Portals of Entry of infection:

Pharyngeal lymphoid tissue like tonsils ‘Bovine type’.

Secondary to pulmonary tuberculosis.

Hematogenous-rare

Tuberculous lymphadenitis is caused by *Mycobacterium tuberculosis*. The basic pathology is a granulomatous inflammation with tubercles which undergoes caseation necrosis and destruction of the lymph node. Spread of infection to the adjacent nodes causing periadenitis results in these nodes getting adherent to each other. This gives the characteristic physical sign of early matting of the node. Where the node lies deep to the deep fascia as in the neck, the caseous node perforates through the deep fascia and the caseous matter escapes into the superficial fascia resulting in the characteristic collar stud abscess.

The condition most commonly affects children and young adults but can occur at any age.

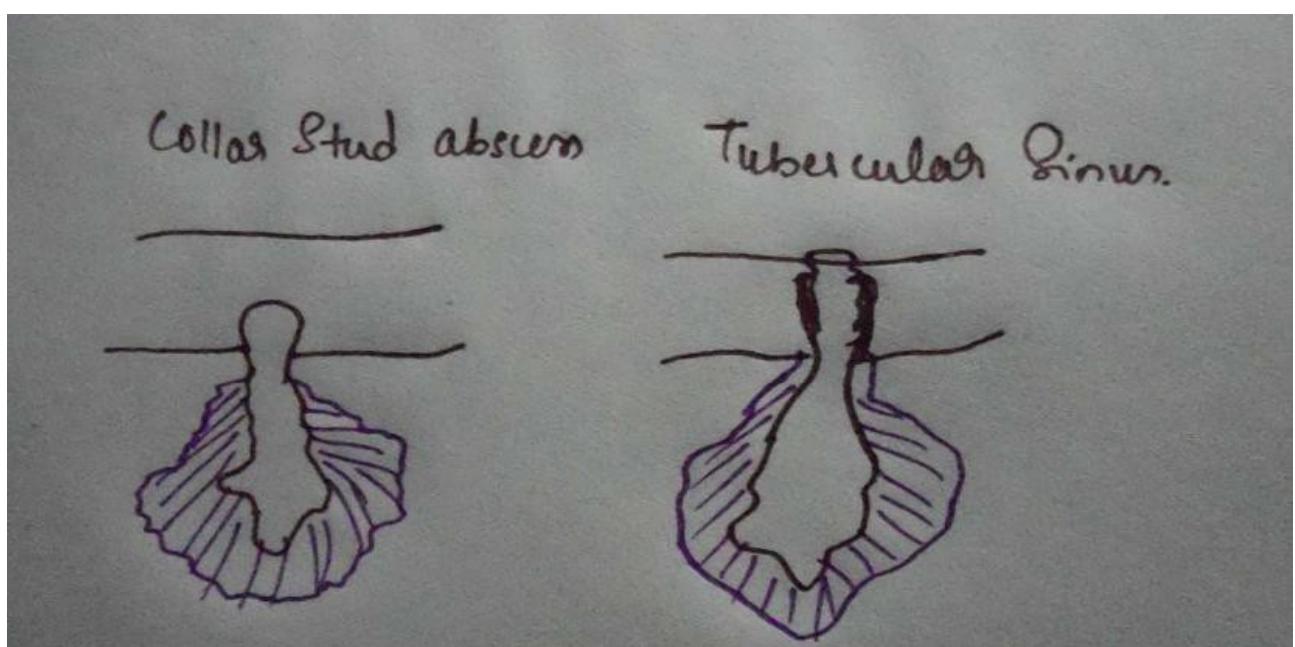
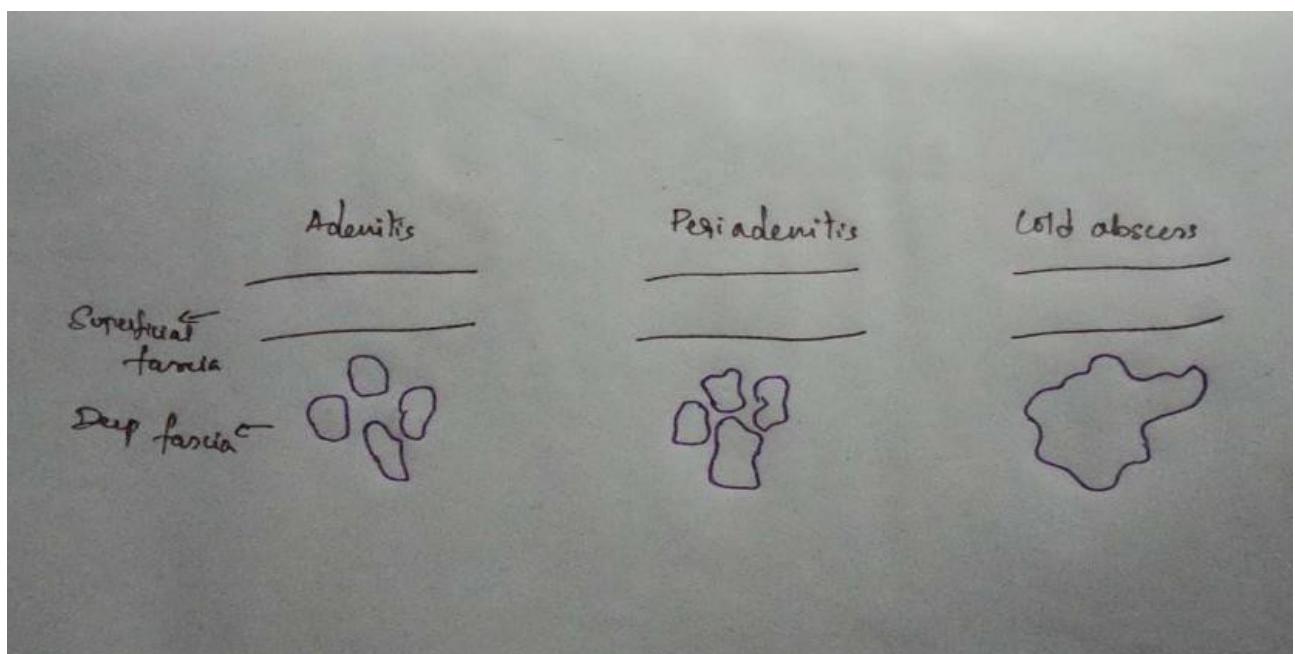
Clinically three stages are recognized².

First stage- The nodes become enlarged without matting. This is known as lymphadenoid type and its differentiation from chronic septic lymphadenitis becomes difficult.

Second stage- In periadenitis the enlarged nodes become adherent to one another (matted). This is the most characteristic feature of tuberculous lymph nodes.

Third stage- caseation takes place in the interior of the nodes so that the nodes become softer with formation of cold abscess. Gradually the cold abscess makes its way towards the skin and ultimately bursts out forming a tubercular ulcer or sinus which refuses to heal.

Pathologically 5 stages of lymphadenitis are recognized:



Stage I – The glands are enlarged, mobile, firm and slightly tender. Histologically this stage shows nonspecific relative hyperplasia.

Stage II – The nodes are enlarged, firm and fixed to surrounding tissue and to each other. Histologically they show peradenitis.

Stage III – The caseation occurs within the lymph node which burst out and collects beneath the deep fascia.

Stage IV – The caseous material perforates the deep fascia and escapes into the superficial fascia resulting in collar stud abscess formation.

Stage V – The cold abscess burst out and gives rise to a persistent discharging sinus.

Case report:

A 38 year old female belonging to lower socio economic class came with chief complaints of left sided swelling in the neck for the past 3 months, Insidious in onset, Progressive in nature and attained its present stage, No Aggravating /Relieving factors. H/o fever on & off for past 3 months, Evening rise of temperature+, No h/o Cough/Hemoptysis/Breathlessness/Loss of weight. No h/o Contact. Rest of the family members were normal and healthy.



Clinical image showing neck swelling

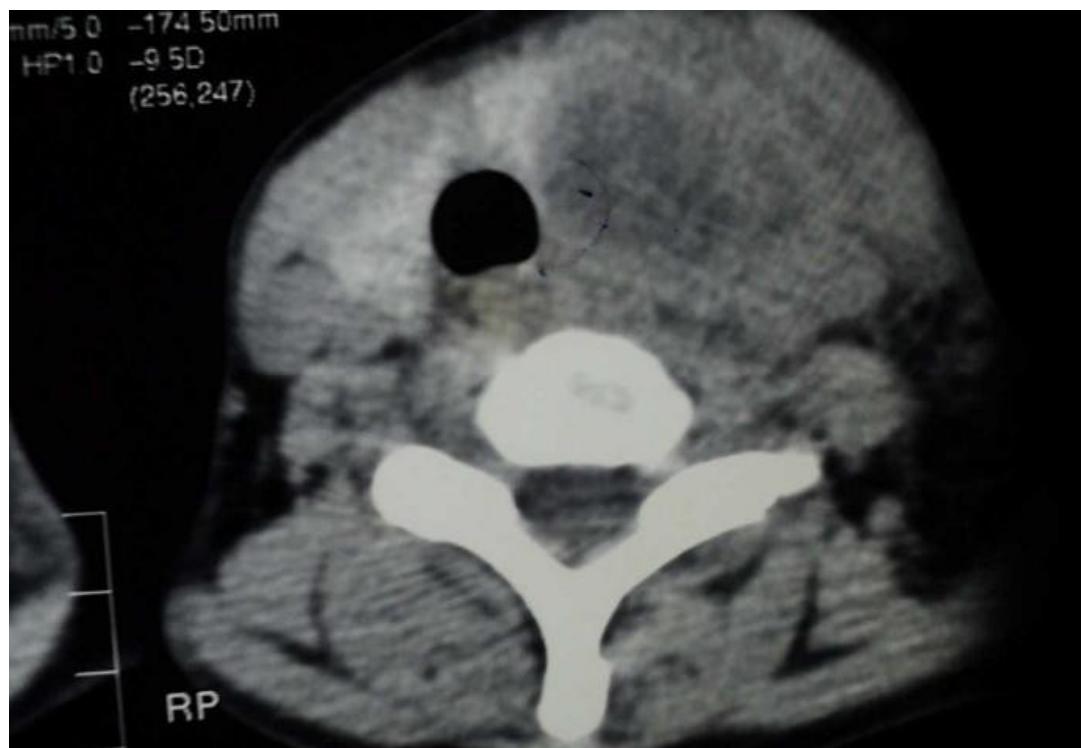
On General examination, patient is moderately built. Pallor+. Temperature-100F, PR-102/min, BP-110/70 mmHg, Respiratory system and other system within normal limit.

Local examination- 5*6cm swelling seen in the left sided anterior neck region, extending up to thyroid cartilage superiorly, 1cm above the sterna notch, Just crossing the midline medially, up to the anterior border of Sternocleidomastoid laterally, Tender on touch, Warmth, No dilated veins, Skin Pinchable, No sinus/scar.

Investigations- Hb-11.2gms%, TC-9300/cmm Neutrophils-25%, Lymphocytes-73%, Eosinophils-2%, ESR-20/30, CXR- NAD, Mantoux – REACTIVE.

FNAC 3

- Granulomatous lesion with epithelial cells. Chest physician opinion obtained-Advice to start ATT Category I.
-



CT NECK IMAGE ^{4,5}

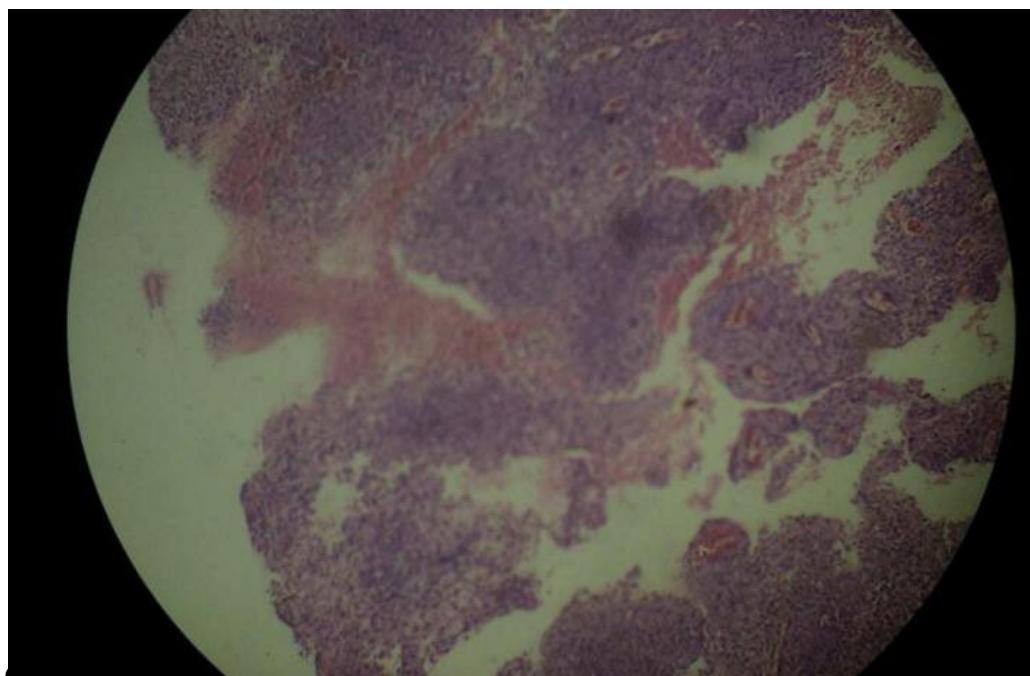


Figure showing histology of the lesion

HPE Report- Granulation tissue with Haemorrhage and Ill defined Epitheloid granuloma, features consistent with partially treated with ATT.

TREATMENT:

Standard anti tuberculosis drugs for six months .our case responded well to 2 RHZ X 4 RH.
UNDER GA, NON DEPENDENT INCISION AND DRAINAGE DONE.

CONCLUSION:

In conclusion, TB is still a common and important disease. It may present in a myriad of ways depending on the organ(s) involved. One should be aware of the various clinical presentations of both pulmonary and extra pulmonary TB. This case is being reported for clinical interest and for the purpose of documentation.

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MUCORMYCOSIS – A CASE SERIES

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Abstract:

Mucormycosis is a rare opportunistic fungal infection with a rapidly progressive and fulminant course with often fatal outcome. A less fulminant variety of this is the Rhino maxillary subtype which usually presents with palatal ulceration, facial swelling, turbinate necrosis and purulent sinusitis. We are reporting our experience of 4 such cases seen during the last 10 yrs. A strong suspicion, prompt diagnosis with pathological confirmation and aggressive surgical treatment gives a better outcome.

Introduction:

Mucormycosis is an invasive fungal infection initiated in the nose and paranasal sinuses that frequently progresses to orbital and brain involvement. If recognized early, involvement is limited to the nasal cavity and paranasal sinuses. The purpose of this article is to report 4 cases of Rhino maxillary variety of mucormycosis, to emphasize the importance of early diagnosis, wide surgical debridement and systemic antifungal therapy in obtaining a favorable outcome.

Case report:

Case 1:

A 62 year old female reported to our department with complaints of pain and swelling in the left cheek for the past 1 month and ulcer in the palate for the past 15 days , gives history of dental extraction done 1 month back., History of swelling around left eye. Patient was a known case of type 2 diabetes mellitus for 2 years and on irregular treatment.

On examination, a diffuse swelling noted in the middle third of face on left side, which was warm and tender. On oral examination, upper lateral incisors were missing. Granulations noted along the alveolar margin. Ulcerative lesion seen in the left half of hard palate of size 3x2 cm and covered with yellowish slough and everted borders. Oroantral fistula seen.

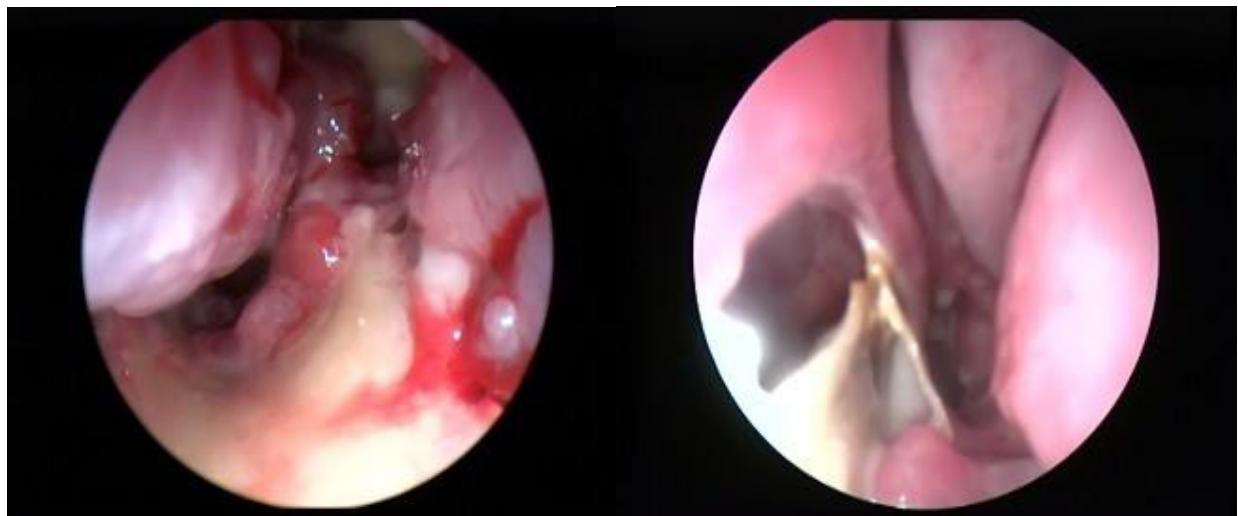
Diagnostic nasal endoscopy was done , pus seen in middle and inferior meatus. Granular mass seen in left nasal cavity and pushing the septum towards opposite side, partly eroding and destroying it. Considering the patient's medical history and a rapidly expanding ulcer involving palate and medial wall of maxilla , a provisional diagnosis of either osteomyelitis maxilla or mucormycosis was made.,even the possibility of malignant growth maxilla was considered .

Patient had the following investigations. Blood sugar – 136 mg/dl, FBS – 180 mg/dl, PPBS – 239 mg/dl, Blood urea – 20mg%, Creatinine – 0.6mgs%. Hemogram showed Hb – 10.4gms%, TC- 6800 cells/mm³ (P – 57%, L – 39%, E – 4%). ESR was 35 and 70mm at ½ hr and 1 hr respectively.



Ulcerative lesion of palate

Granulation tissue in alveolar margin



DNE showing granulations

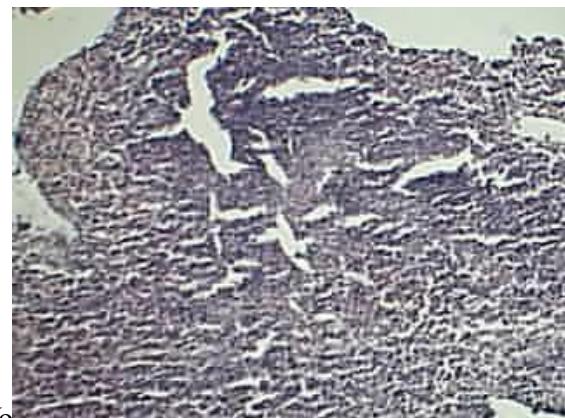
Septal perforation

CT PNS showed diffuse radio- opacity of the left maxillary sinus and left nasal cavity with bony erosion ,palatal destruction with oroantral fistula. Patient was taken up for surgery and,wide Surgical debridement done including removal of part of the alveolar margin and medial wall of maxilla ., wide middle meatal antrostomy created. Necrotic material and granulation tissue removed from maxillary sinus.. The materials were sent for histopathological examination, which was reported as mucormycosis (Path No.1016/09). Patient started to show improvement from next day. Subsequently after HPE results, patient started on antifungal treatment (Tab. Itraconazole 100mg bd for 1 month) and broad spectrum antibiotics (Inj. Ceftriaxone 1gm iv bd for 15 days and Inj. Gentamycin 80mg iv bd for 1 week). Patient's diabetes controlled with Insulin. After 2 weeks post operatively, cheek swelling improved. Palatal lesion healing well. Normal lining of nasal mucosa started to appear. DNE done 3 weeks Post OP. Wound healing well, no granulations, antrostomy functioning well and oral antral fistula closed. Patient discharged and advised to control diabetes mellitus with Insulin treatment. Is disease free for past 2 yrs.



Pre op cheek swelling

Post op picture 2 weeks later



Post op alveolar region

Histopathology picture

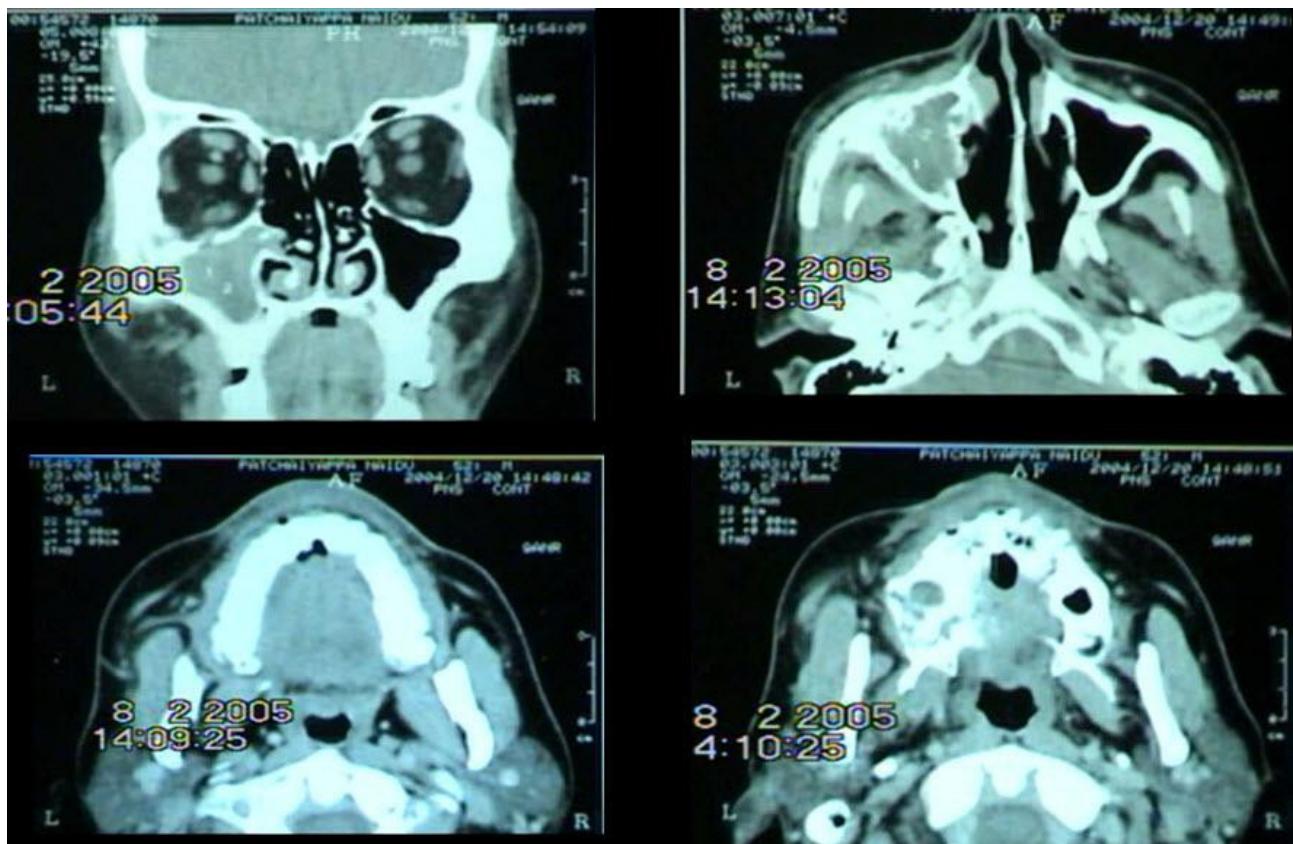
Case 2:

A 52 yr old male presented with midline palatal ulcer 1 month duration, gives H/O swelling in midline palate with history of needle aspiration done elsewhere. He is a known Diabetic on irregular treatment and hypertensive. Clinical examination revealed 5 x 3 cm oval shaped ulcer in the palate more on left side exposing the bone. Left side cheek swelling and Paraesthesia present.

Investigations revealed blood sugar 220 mg/dl. CT revealed opaque left maxillary sinus with palatal erosion. Biopsy reported as granulation tissue with fungal elements. Proceeded with partial maxillectomy /surgical debridement. HPE proved Mucormycosis. Itraconazole was given for 3 wks and on adequate control of disease same was stopped. Wound healed well, pt was given an obturator and is disease free for past 6 yrs.



Palatal ulcer with necrosis of alveolar margins



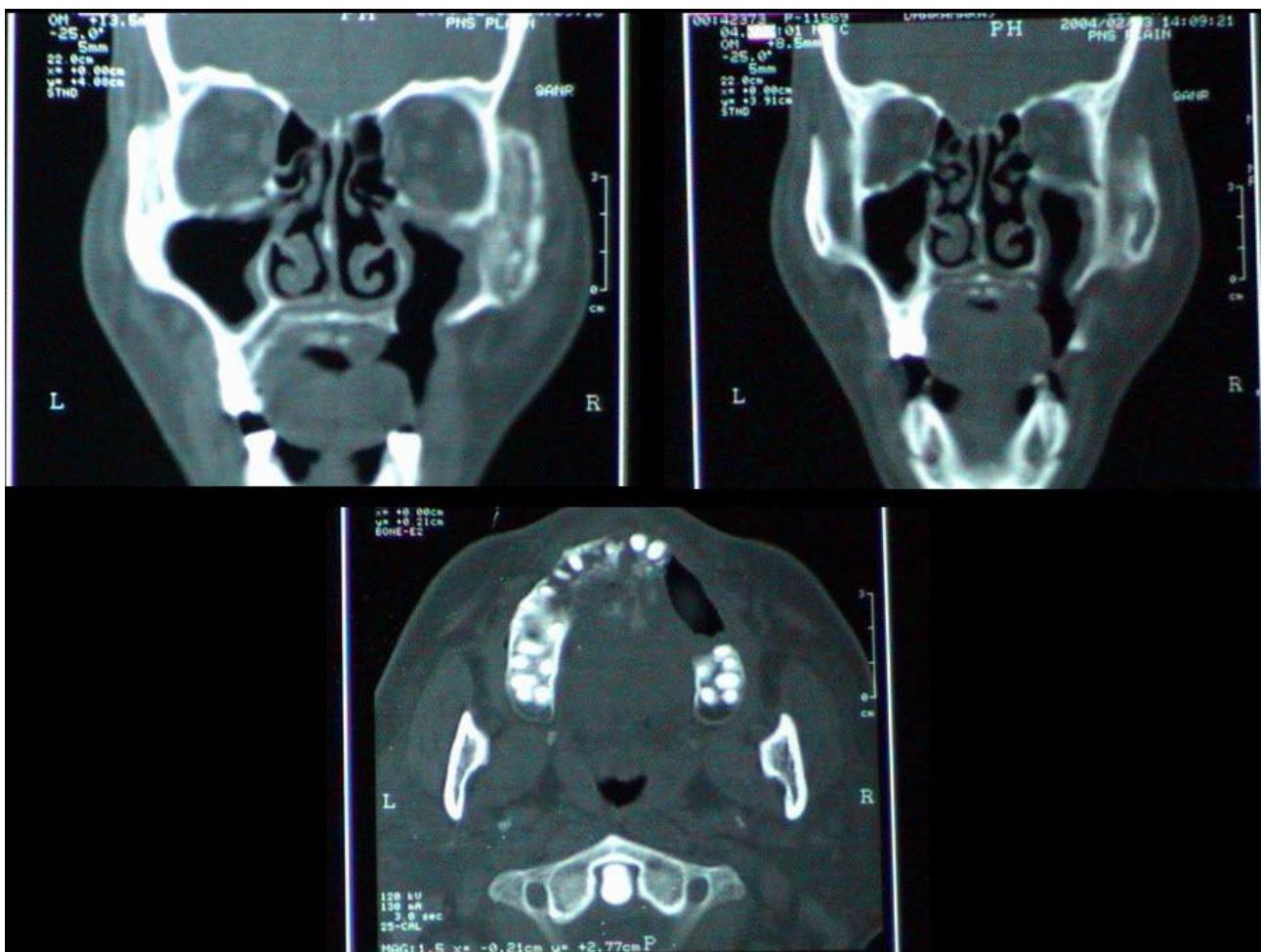
CT Images



Post op picture with obturator

Case 3:

A 50 yr old male presented with swelling and numbness over the rt cheek and upper lip for 6 months, mucopurulent discharge over the alveolar margin for 5 months. Gives H/O dental extraction of 3 tooth, and says he developed this problem following it. On clinical examination,mucopurulent discharge noted in floor of rt nasal cavity, X ray PNS showed bilateral maxillary sinusitis, CT revealed oroantral fistula on rt side. Biopsy reported as sub acute sinusitis with osteomyelitis with evidence of mucormycosis. Control of diabetes initiated and wide surgical debridement with sequestrectomy of devitalized floor of maxilla done.HPE confirmed mucormycosis. Pt was given obturator on dental opinion and is disease free .



CT scan showing oroantral fistula



Defect seen after removal of the disease

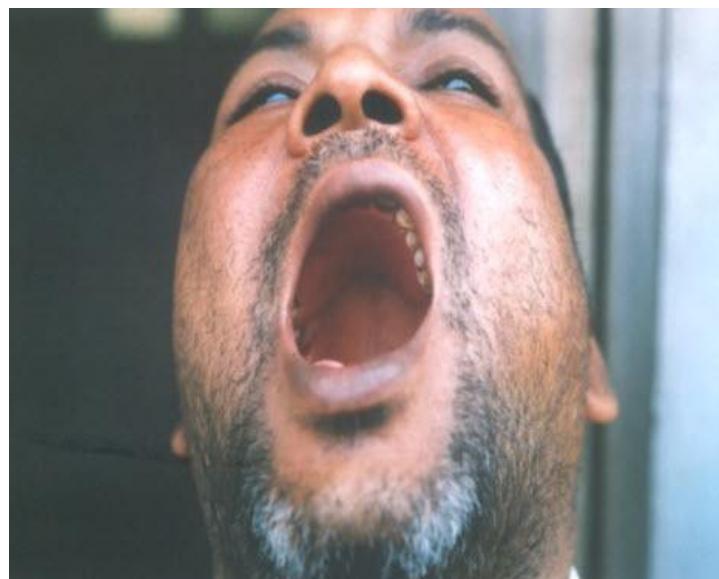
Case 4:

A 53 yr old male presented with ulcer in the palate for 1 yr. He had uncontrolled Diabetes.,gives H/O dental extraction. Clinical examination revealed a large ulcerated lesion over the rt alveolar margin extending to the palate .CT revealed oroantral fistula.Surgical debridement done,almost the entire floor of rt maxilla was unhealthy and was removed as a sequestrum.HPE of the specimen confirmed mucormycosis. This patient was better during the post op period,,was having problems with the interim obturator and was being attended by the dental department., was lost to our follow up . It was later known that this patient expired due to diabetes related renal complications.

PRE OP



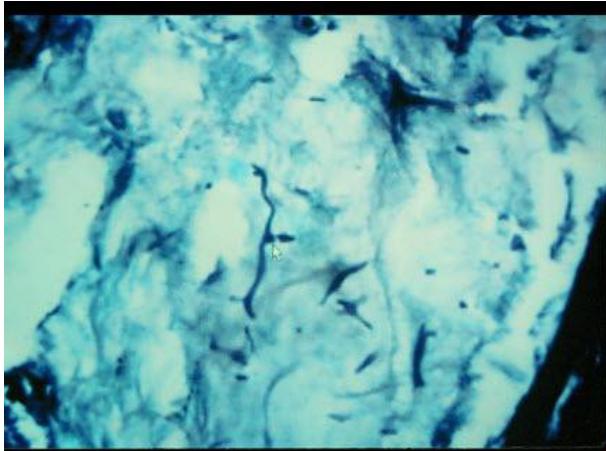
Figure showing palatal defect



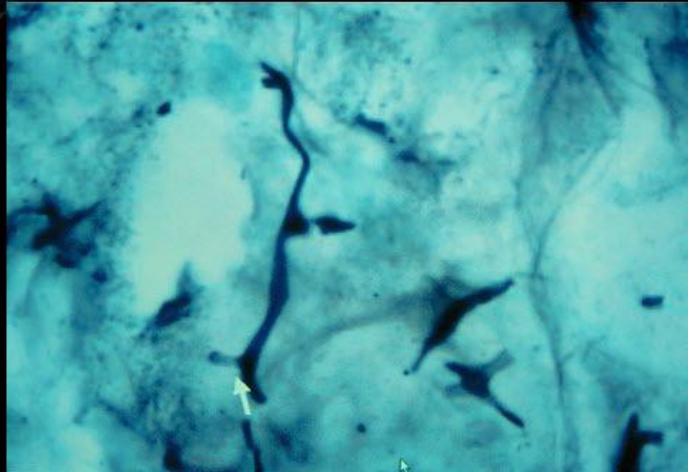
Patient with obturator



Pre op picture and patient with obturator



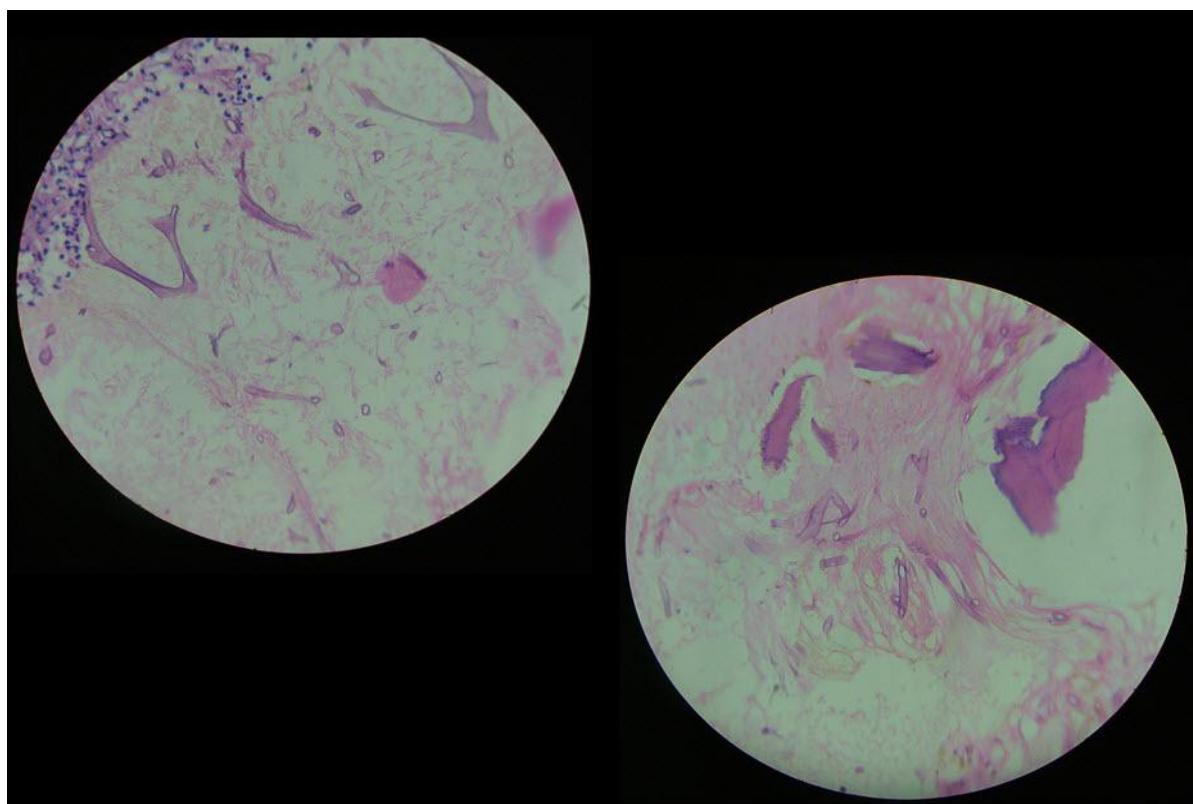
Grocott-Gomori
methanamine-silver stain



Discussion:

Mucor mycosis is a rapidly progressive opportunistic infection caused by an organism of the family mucoraceae order mucorales class zygomycetes. These fungi are ubiquitous found in soil, bread molds, decaying fruits and vegetables, usually are harmless and become pathogenic in man under certain conditions like immunosuppression, diabetic acidosis, antibiotic, steroids and cytotoxic therapy, with other predisposing factors like malignancy, burns, malnutrition, renal failure and blood dyscrasias. The affinity for Diabetic patients is related to the fact that Rhizopus organisms thrive in an environment rich in glucose and an acid PH, because they have an active ketone reductase system, decreased phagocytic activity of polymorphonuclear leucocytes may also be a factor¹.

Upper airway mucormycosis was first described in 1885 by Paltauf, who coined the term Mycoris mucorina, which subsequently became Mucormycosis⁽²⁾⁽⁷⁾. Mucormycosis can infect the lungs, central Nervous System, Gastro intestinal tract and skin (burns patients) but is known best for its rhinocerebral presentation, which is usually initiated with sinonasal involvement and may progress to the orbit and the brain². Rhinocerebral form is further subdivided into two subtypes : a highly fatal rhinoorbitocerebral form which is invasive and may involve the ophthalmic and internal carotid arteries and a less fatal rhinomaxillary form which involves the sphenopalatine and greater palatine arteries, resulting in thrombosis of the turbinate and necrosis of the palate³. All our four cases belong to this group.



Histopathological image showing fungi with aseptate hyphae

Everyone is exposed to and inhales spores of the mucorales; the nasal ciliary clearance system, however transports these spores out of the nasal cavity, down the pharynx, to be cleared out by the gastrointestinal tract. Spores inhaled into the lungs are cleared by phagocytes. In the susceptible individual, the infection usually begins along the middle or inferior turbinate². Depending on the degree of immunocompromise the disease process can be indolent or fulminant. Once the spores have entered the tissues, the organism becomes angioinvasive and has a predilection for the internal elastic lamina of the arteries. This invasion causes thrombosis, with secondary ischemic infarction and haemorrhagic necrosis. The organism thrives in the necrotic tissue and spreads by direct extension along injured blood vessels¹.

MEDICALLY IMPORTANT MEMBERS OF THE CLASS ZYgomycetes²

| Class | Order | Family | Genus | Species |
|------------------|-----------|--------------------|--------------------------|-----------|
| Zygomycetes | Mucorales | Mucoraceae | Absidia* | |
| | | | Mucor* | |
| | | | Rhizomucor* | |
| | | | Rhizopus* | |
| oryzae(or | | | | |
| bertholetiae | | Cunninghamellaceae | Cunninghamella* | Arrhizus) |
| | | Mortierellaceae | Mortierella* | wolfii |
| | | Saksenaeacea | Saksenaea* | |
| | | Syncephalastraceae | Syncephalastrum elegans* | |
| | | Apophysomyceae | | |
| | | Thamnidiaeae | Cokeromyces | |
| Entomophthorales | | | | |
| | | Ancylistaceae | Delacroixia | |
| | | | Basidiobolus | |

*Reported to have caused rhinocerebral mucormycosis.

Involvement of the oral cavity usually appears as palatal ulceration or necrosis and later as perforation of the palate as a result of infection in the nasal cavity or paranasal sinuses. Patients often exhibit facial cellulitis and anaesthesia, nasal discharge, necrotic turbinates, fever, headache and lethargy³. These findings were noted in our patients.

Early and late cavernous-carotid fistulas and mycotic aneurysms of the carotid have been reported. Patients should therefore be followed for sometime after the initial response to therapy¹.

Differential diagnosis of the lesion should include Squamous cell carcinoma, Chronic granulomatous infection like tuberculosis, tertiary syphilis, midline lethal granuloma and other deep fungal infections.

Radiographic analysis by routine radiographs may reveal clouding of multiple sinuses, mucosal thickening and bone erosion. CT better defines soft tissue invasion and necrosis, early bone erosion and cavernous sinus thrombosis. All our patients exhibited these findings with bone erosion and

oroantral fistula. MR imaging, with or without gadolinium, is the best way of evaluating changes in major vessels, including carotid artery thrombosis and cavernous sinus thrombosis, and any intracranial extension.

Once patient presents with above said findings, tissue diagnosis should be established with a biopsy to look for non septate hyphae representing fungal invasion. The best demonstration of the tissue invasive, nonseptate hyphae (cellophane tubules) is with silver methanamine stain¹.

As the disease progresses with alarming rapidity, early diagnosis, prompt and aggressive therapy is essential⁴. Successful treatment of mucormycosis consists of aggressive repeated surgical debridement of necrotic tissue, systemic antifungal therapy and immediate control of underlying systemic diseases. Correction of acidosis and hyperglycemia contributes to better survival rates. The fungus thrives in devitalized and necrotic tissue hence rigorous debridement is indicated¹. Areas of ischemic tissue should also be removed because vascular thrombosis prevents chemotherapeutic agents from reaching the diseased tissues.

In acute fulminant fungal sinusitis with invasion of blood vessels, amphotericin B has been considered the drug of choice(at a dose of 1 – 1.5 mg/kg per day). There is now growing consensus that the newer lipid based formulations of the drug,in particular high doses of liposomal amphotericin B (10-15 mg/kg per day),should be administered and be continued until the patient recovers⁵.Renal parameters have to be continually monitored.Other agents like Itraconazole, Voriconazole and Posaconazole have been reportedly tried with varying results., however, the optimum dosage and duration of treatment have not been defined⁵. Itraconazole (100 mg bd) results in remineralization of the eroded skull base⁵ and is also said to prevent recurrence.The role of topical amphotericin B has not been studied systematically,but because of the small risk associated with administration it is often employed.(50 mg vial of intravenous amphotericin B and 10 ml of sterile water, in a dosage of 4 ml in each nostril 2 to 6 times daily). This can also be nebulized into the nose with a Rhinoflow device². Hyperbaric oxygen therapy is theoretically attractive because it reverses the ischemic acidotic conditions that perpetuate fungal growth. Hyperbaric oxygen treatments are usually given at two atmospheres for 1 hour on a daily basis for upto 30 treatments. This may limit deformity by decreasing the required area of debridement⁶.

Conclusion:

Early diagnosis is important in the success of treatment. A patient presenting with nasal and palatal lesions, facial swelling or cellulitis with history of dental extraction and being a known diabetic should immediately rise a suspicion of this dreadfull condition. Active surgical treatment with removal of all dead and necrotic tissue, control of diabetes and systemic antifungal therapy gives good results,reducing the mortality and morbidity due to mucormycosis.

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Letters to the Editor

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To
The Editor,
Online journal of Otolaryngology

Dear Sir,

The review article “ BIPP (Bismuth Iodine Paraffine Paste) Pack revisited,” by Dr T. Balasubramanian sheds light on BIPP packing. Used extensively in pre-penicillin era, this armamentarium in the hands of the otolaryngologist is almost obsolete.

It has the following disadvantages, as a result of which it's use has been discontinued :

- 1)Toxicity to bismuth¹
- 2)Toxicity to iodoform²
- 3)Increased pain, both due to BIPP nasal pack and during pack removal³
- 4)Septal perforation³ following BIPP pack
- 5)Vestibulitis³ following BIPP pack
- 6)Unpleasant smell⁴

However, BIPP is still in use at some centres. Paulose⁴, has mentioned on his website that he uses BIPP packing after mastoidectomy.

An alternate formulation, Whitehead's varnish, an iodoform compound, is a safer alternative to B.I.P.P. gauze.²

The constituents of Whitehead's varnish are⁵:

- (i) Iodoform 10 gm
- (ii) benzoin 10 gm
- (iii) Starch 7.5 gm
- (iv) Natural balsams 5 gm
- (v) Solvent ether to 100 ml.

Iodoform has an anaesthetic action on mucosal surfaces and this contributes to amelioration of pains. Also, its application to tissues leads to slow release of elemental iodine which exerts a disinfectant effects.

Cascarini⁶ has stated that Whitehead's varnish has many applications, which includes packing of the nose and in the mouth. Its constituent aromatic resins break down to produce benzoic acid, which is an antiseptic. In addition, its waterproofing property confers an advantage.

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